INTENSIVE VOICE TREATMENT FOR CHILDREN WITH SPASTIC CEREBRAL PALSY

by

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ABSTRACT

Purpose: The purpose of this study was to examine the effects of an intensive speech treatment for children with spastic cerebral palsy (CP).

Background: Children with spastic CP have speech and voice disorders, which may limit functional communication and negatively impact quality of life. There are limited published outcome data on speech treatment approaches for these children. Recent advances in theories of motor development and behavioral gait and limb treatment provide a solid framework (consistent with theories of motor learning) from which to test different speech treatment concepts (e.g., intensive treatment, high effort exercises, repeated practice trials, and sensory augmentation/sensory awareness training) in children with spastic CP.

Method: A multiple baseline single-subject design with replication across participants (five children with spastic CP) was used. Acoustic measures related to voice functioning, auditory-perceptual analysis of speech samples, and perceptual ratings by parents of participants in this study were obtained from baseline, post-treatment, and 6 week follow-up data recording sessions.

Results: The four participants who received treatment demonstrated a marked change in performance on one or more of the acoustic measures and there were strong listener preferences for the treated speech samples (post-treatment or follow-up sessions) over baseline samples for most perceptual characteristics rated. In addition, parents of these four participants reported improved perceptions on two or more voice, speech, or communication characteristics following treatment, and all had an overall favorable impression of their child's treatment outcome and of the treatment approach used. No changes were observed in the one participant with CP who did not receive treatment. *Conclusions:* These findings suggest that in these four participants with CP, intensive speech treatment changed the output of the speech motor system in a manner that listeners preferred over baseline speech samples. In addition, this positive effect was maintained in nearly all cases 6 weeks after the conclusion of treatment. These findings are consistent with those in limb and gait treatment literature, thus highlighting potential key treatment concepts to consider in behavioral treatment for children with spastic CP.

INTRODUCTION

Overview of Cerebral Palsy

Cerebral palsy (CP) is the most common movement disorder in children. The definition of CP is "...an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development" (Mutch, Alberman, Hagberg, Kodama, & Perat, 1992). Because of its varied etiology and course of development, CP is often defined by clinical description without information related to etiology, pathology or prognosis of the condition (Scherzer, 2001). Accordingly, the age of diagnosis of CP ranges from as early as several months after birth for severe cases, to as late as five years of age in mild cases (Stanley, Blair, & Alberman, 2000). The prevalence of CP in industrialized countries is 2 to 2.5 cases per 1000 live births (Gupta, 2001; Rosen & Dickinson, 1992). This prevalence has been increasing in industrialized countries since the mid-1970s, a trend that may be partially attributed to the increase in survival rate of very low birth weight infants. To understand the diverse nature of CP, it is necessary to examine its etiology, classification and descriptive terminology, and associated impairments.

CP can be caused by congenital or acquired etiologies. Congenital CP accounts for approximately 85% of cases and is the result of damage or dysfunction during the prenatal, natal, or postnatal time (Gupta, 2001). Premature birth is one major cause of congenital CP. Approximately 40% of children with CP were either born prematurely, or had a low (<2500 g) or very low (<1500 g) birth weight (Gupta, 2001). Damage to the white matter in the brain, which is collectively called perinatal leukoencephalopathy, is the major cause of abnormal neurodevelopmental outcomes related to preterm birth. Other causes of congenital CP include infants who suffer birth asphyxia during the delivery process, hyperbilirubinemia (related to Rh factor and other blood group incompatibilities), and a variety of prenatal factors, such as exposure to toxins, maternal infections, and malformations of the central nervous system (hydrocephalus or encephalocele). Acquired CP accounts for approximately 15% of all cases and is the result of factors that occur in the early developing years of an infant or child (Pearlstein, 1952). Causes of acquired CP may include infections (e.g. meningitis, encephalitis), trauma (e.g. motor vehicle accidents, shaken baby syndrome), and other factors such as inadequately treated hydrocephalous.

CP is classified into various types (Scherzer, 2001; Stanley et al., 2000). Classification of CP is based heavily upon clinical description of the neurological signs rather than etiology or neuroanatomical localization of a lesion; however, lesion localization may correlate with the observed motor signs. Spastic CP is the most common type of CP and is the focus of this paper (Brunstrom, 2001; Rosen &Dickinson, 1992). Spastic CP generally correlates with a lesion (or lesions) in the direct motor pathway including the motor cortex and its descending white matter tracts. The strictest definition of spasticity is a velocity-dependent increased resistance to passive movement (Fowler, Ho, Nwigwe, & Dorey, 2001; Thompson, Parmer, Reier, Wang, & Bose, 2001). An alternative and expanded meaning of the term spasticity includes a constellation of motor signs such as increased tone, hyperactive reflexes, weakness, and poor coordination (Goldstein, 2001; Thompson et al., 2001). For the purpose of this discussion, the term spasticity will be limited to its strictest definition of a velocity dependent increased resistance to passive movement. Other motor signs that are included in the umbrella definition of spasticity will be described separately as they occur in spastic CP.

The motor impairment in children with spastic CP can be characterized as having both positive and negative signs (Goldstein, 2001). The positive motor signs include spasticity, increased tone, increased deep tendon reflexes including increased amplitude and spread of the reflex reaction, persistent and exaggerated primitive reflexes, clonus, Babinski sign, and discordant mass activation of muscles (Goldstein, 2001; Hadders-Algra, 2000a). Negative motor signs include decreased coordination through loss of selective muscle control, decreased speed of movement, decreased strength, and decreased endurance (Goldstein, 2001). The distribution of motor signs associated with CP varies and is classified according to limb involvement. Common terminology includes: *monoplegia* - involving one extremity; *hemiplegia* - involving both upper and lower extremities on 1 side of the body; *paraplegia* - involving both lower extremities; *diplegia* - involving all four extremities with only mild involvement of the upper extremities; and *quadriplegia* - equal involvement of upper and lower extremities. Although motor signs are most commonly discussed in relation to limb and body movements, these signs can negatively impact the speech of children with spastic CP as well.

Speech disorders have been reported to occur in children with spastic CP (Clement & Twitchell, 1959; Solomon & Charron, 1998; Workinger & Kent, 1991). The most common perceptual characteristics include consistent hypernasality, breathy voice quality and voice quality changes (Workinger and Kent, 1991), monotonous speech, reduced loudness, and uncontrolled rate and rhythm of voice (Clement and Twitchell, 1959; Heltman and Peacher, 1943; Keesee, 1976; Wolf, 1950). Disordered respiration, including short vowel durations, shallow inspirations with forced expirations, and weak respiratory muscles have been reported (Clement & Twitchell, 1959; Solomon & Charron, 1998; Wolfe, 1950). In addition, disordered articulation, including difficulty in producing lingual/dental sounds, in raising the velum for plosive sounds, pursing the lips, and difficulty with articulation requiring back tongue movements, have been reported in these children (Hixon & Hardy, 1964, Clement & Twitchell, 1959; Wolf, 1950). Furthermore, children with spastic CP are at risk for progressively worsening of speech breathing and phonatory problems as they mature (Keesee, 1976; Workinger & Kent, 1991).

Although the motor signs (limb and speech) are predominant in children with spastic CP, there are a variety of other associated deficits that can occur. These deficits include visual impairments, hearing impairments, language disorders, seizure disorders (most common in hemiplegia, and quadraplegia), learning disabilities, social and emotional problems, and mental retardation (most often associated with severe CP, more common in quadraplegia) (Brunstrom, 2001). To understand how motor signs evolve and impact functional abilities of children with spastic CP, an overview of motor development in this population is discussed below. This is followed by a discussion of treatment for these motor signs in limb, gait, and speech of children with spastic CP.

Motor Development in Children with Spastic CP

Motor signs in infants and children with CP emerge over time and are not static (Twitchell, 1965). During development, motor signs such as increased tone and spasticity may manifest as late as 7 to 9 months of age, eventually leading to the diagnosis of CP (Scherzer, 2001). In addition to emerging abnormal motor signs, most children with CP are delayed in achieving developmental milestones. For example, the typically developing infant exhibits anticipatory postural control and begins to scale muscle activity to task conditions during reaching movements by 15 months, whereas infants with CP may not develop these skills until 18 months of age or later (Hadders-Algra, van der Fits, Stremmelaar, & Touwen, 1999). Similarly, speech and language development is often delayed in children with CP who may not speak their first word until 15 months of age or later (Byrne, 1959).

The neural processes that underlie delayed and disordered neuromotor development in children with CP are not fully understood. Theories of motor development in typically developing children are often applied to children with CP in an attempt to understand motor development in a damaged nervous system. Three theories of motor development are briefly reviewed here including the *neural-maturationist theory*, *dynamic systems theory*, and *neuronal group selection theory*. Although these theories have limitations in explaining both normal and disordered development, they provide a theoretical framework from which to explore plausible treatment strategies.

The *neural-maturationist theory* contends that motor development is based on a gradual unfolding of pre-determined patterns in the central nervous system. As a child develops, there is increased cortical control over primitive reflexes. This theory places the primary and almost exclusive driving force for maturation within the nervous system (McGraw, 1943). For example, newborn stepping is a behavior that occurs when an infant is held upright with the trunk bent slightly forward and the feet touching a stable surface. This rhythmical stepping behavior typically disappears at about 4 to 6 weeks of age, then reappears late in the first year of life just prior to intentional stepping for walking. The neural-maturationist theory explains that newborn stepping is a reflexive behavior that will disappear as the cortex matures and exhibits control over primitive reflexes. The reappearance of stepping is facilitated by a different and higher level of cortical control for voluntary movement (McGraw, 1943). Thus, neural-maturationist theory asserts that changes in behavior reflect changes in the brain, with relatively little consideration of the influences of environment, motivation, and task constraints on movement (Hadders-Algra, 2000b; Thelen, 1995).

Detection of motor dysfunction in light of this theory is based on late attainment of motor milestones and the presence of abnormalities in muscle tone and reflexes in infants and children with CP. Treatment for children with spastic CP based upon this theory focuses on normalization of muscle tone, and facilitation of normal movement patterns without unwanted increased muscle activity. Although neural-maturationist theories have dominated developmental motor literature for many years, new theories have emerged that challenge many of its concepts. In short, it has become evident that the simple unfolding of motor programs through maturation of the nervous system is inadequate to explain all the nuances of normal and disordered neuromotor development (Hadders-Algra, 2000a; 2000b).

In the early 1980s, an alternative to the neural-maturationist theory came about with *dynamic systems theory*. Dynamic systems theory is based upon a combination of the work of Bernstein (1967) in motor control and principles of nonequilibrium thermodynamics (Thelen, 1995), and has been applied to human motor development by Thelen and colleagues (Kamm, Thelen, & Jensen, 1990; Thelen, 1995; Thelen & Fisher, 1983). Thelen (1995) described development in dynamic systems theory as a process of self-organization, brought about by the interaction of multiple factors, such as body weight, muscle strength, joint configuration, the infant's mood, specific environmental conditions and brain development. In contrast to neural-maturationist theory where the nervous system is the primary driving force in development, no one system (e.g. neural, muscular, environmental) has a priority over another in dynamic systems theory. Movement is an emergent factor of them all.

Consider again newborn stepping. The neural-maturationist theory described it as a primitive reflex that disappeared with cortical development. Thelen and Fisher (1983) would argue that factors other than, or in addition to, the nervous system contributed to the "disappearance" of newborn stepping. In a series of experiments, Thelen and colleagues (Kamm, et al., 1990; Thelen, 1995) demonstrated that: (a) newborn stepping and newborn/infant kicking behavior appeared isomorphic on kinematic analysis suggesting that this pattern had not disappeared, but was influenced by gravitational forces and sensitive to postural changes, (b) newborn stepping could be inhibited by placing weights on the newborn's legs, which simulated weight gain that typically coincides with the developmental disappearance of newborn stepping, and (c) newborn stepping could be elicited after it had "disappeared" by placing a newborn in warm water, which eliminated the influence of developmental weight gain (increased load) on the infant's legs. The authors concluded that the disappearance of newborn stepping might be due to biomechanical effects of increased weight gain and gravitational factors for which infants are too muscularly weak to overcome. The reappearance of stepping behavior later in the first year of life may be a combination of increased muscular strength to lift the legs and a motivation to locomote in the environment. Thus, the patterns of infant leg movement, including newborn stepping and newborn/infant kicking, emerge from the multiple influences of neuromuscular activation, arousal, and changing forces (weight and gravity) that act on the legs as they move (Kamm et al., 1990).

An important concept in the application of dynamic systems theory to motor development is that of an attractor. An attractor is a preferred, but not mandatory, configuration that the system easily falls into, or prefers to fall into, even after interruptions or perturbations (Kamm et al., 1990; Thelen, 1995). Thelen describes attractors as having wells of varying depth (see Figure 1). Stable motor patterns are represented by deep attractor wells, which are stereotypical, almost obligatory patterns. Less stable motor behaviors represent shallower attractor wells, which are preferred, but flexible patterns that are more sensitive to perturbation. Development, then, is described as "... a series of states of stability, instability, and phase shifts in the attractor landscape, reflecting the probability that a pattern will emerge under particular constraints" (Thelen, 1995, p.84). Accordingly, any attractor or motor pattern that is too stable (well too deep) can interfere with developmental progress. Dynamic systems theory dictates that the system must be able to explore beyond its current state, both perceptually and motorically, in order to move into a new phase of motor development. Children with spastic CP may be unable to adequately explore their environment which limits phase shifts (periods of instability) and development of new, more functional attractor states. As a result, children with CP are at risk for getting "stuck" in attractor states that may be appropriate for one phase of neural, motor, or emotional development, but are not functional or adaptable in another.

Given the influence of multiple factors on motor development in dynamic systems theory, there are a variety of opportunities for modifying motor behavior through environmental manipulation (Kamm et al., 1990; Hadders-Algra, 2000b). First, Kamm et al. (1990) expressed the importance of treating the system early to capitalize on neural plasticity. Second, the best time for intervention is when the system is in a phase shift, such as adapting to a growth spurt, or a new emotional or motivational influence. Third, many children with spastic CP will have poor patterns of behavior (deep attractors) already developed by the time they are seen for behavioral treatment. In this case, the goal of treatment is to disrupt the current stability in the system (perturb the system), facilitate experiences that allow for exploration of multiple sensorimotor options, and shift behavior into a more functional stable motor pattern through practice (Kamm et al., 1990). Perturbation in this sense is not an immediate shift away from and return to the current state of motor functioning; rather, it is a shift from one state of motor functioning into a completely different (potentially improved) state of motor functioning.

Although dynamic systems theory recognizes the nervous system as a contributing factor to emerging motor behavior, one criticism has been that it does not the define the specific neural mechanisms of developing or mature motor systems or the consequences of atypical brain development (Sporns & Edelman, 1993). In contrast, a third prominent theory of motor development, called the *neuronal group selection theory* (NGST), addresses specific neural mechanisms and is explicitly based on principles of neuroanatomy and neurophysiology (Edelman, 1978; 1993; Sporns & Edelman, 1993). At the same time, NGST shares many concepts with dynamic systems theory, such as recognizing the importance of nonlinear self-organizing interactions within a complex system (Sporns and Edelman, 1993).

In NGST, the initial phase of neuromotor development involves primary repertoires, which are cortical and subcortical cells that are grossly connected and largely unstructured in the nervous system (Edelman, 1978; 1993; Sporns & Edelman, 1993). These primary repertoires are explored through self-generated movements during fetal life and infancy in a phase called primary variability. Although movements in this phase are variable, the variability is not random; rather, variation is set forth by genetic information that has evolved over time (Edelman, 1993). The self-generated exploratory motor activity in primary variability is not strictly tuned to environmental conditions. This spontaneous and exploratory motor activity allows the system to discover all possible motor solutions (set forth by genetics) for a given goal.

From the exploration of primary variability, somatic selection begins. Somatic selection is the competitive strengthening of neural connections (synaptic connections) involved in the generation of successful movements for a given motor behavior. In order to select movements that are successful, "... the organism must rely both on spontaneous (though not necessarily random) motor activity and the subsequent selection of those movements that simultaneously match environmental demand and internal value" (Sporns & Edelman, 1993, p. 970). Thus, NGST integrates both the specifics of the nervous system and the influence of contributing factors such as the environment and internal state. The timing and duration of somatic selection is behavior specific. For example, sucking patterns may be well-selected at the time of birth, whereas, walking patterns are not selected until the after the first year of life (Hadders-Algra, 2000a).

Secondary neural networks, which are represented as large collections of parallel channels in the nervous system, are created based on continued experience with motor behaviors (Edelman, 1993; Sporns & Edelman, 1993). This phase is called adaptive secondary variability which begins in early childhood and lasts for many years, maturing in late adolescence (Hadders-Algra, 2000a). Practice that occurs during this phase of development refines motor behavior. In the mature adult, the system has the ability to adapt movements precisely and efficiently to task-specific conditions when there are imposed task constraints, or the system can use multiple motor solutions or strategies to accomplish a single motor task when there are no imposed constraints (Hadders-Algra, 2000a).

In infants and children with CP, it is hypothesized that primary repertoires are reduced due to nervous system damage (Hadders-Algra, 2000a, 2000b; Hadders-Algra et al., 1999). Reduced neural circuitry limits self-generated exploratory motor behavior and accompanying afferent information characteristic of primary variability. Selection in infants and children with spastic CP may be impaired for two reasons: (a) decreased primary repertoires offer limited neuronal circuitry from which to select, and (b) decreased exploration offers limited experience from which to select the most successful motor strategies. Limited sensorimotor experiences in children with CP (related to physical motor impairment) result in restricted opportunities for practice and exploration of selected motor patterns. This manifests as decreased motor adaptability, flexibility and refinement in motor behavior of children with spastic CP that normally develops during the phase of adaptive secondary variability.

One goal of treatment in light of NGST may be to facilitate self-generated movements through positioning, posturing, enhancing sensory experiences, and arousal. These variable experiences in a highly plastic, albeit damaged, nervous system may enhance primary repertoires. Providing an infant with an increased number of trial and error experiences (more than a typically developing infant requires) may facilitate the process of selection of successful and functional motor patterns. Finally, children with spastic CP may require active and ample practice of selected motor patterns in a variety of environmental and task conditions to develop adaptive motor behavior and functional secondary repertoires (Hadders-Algra, 2000b).

In summary, motor development in children with spastic CP can be considered in light of several theories of normal motor development. The neural-maturationist theory views motor development as a simple reflection of the maturation of the nervous system. Treatment based on this theory attempts to confine movements to within a normal range, thus potentially restricting sensorimotor experiences and exploratory behavior. Dynamic systems theory views motor development as emerging from multiple and equally contributing systems including neural, muscular, emotional, and environmental systems. NGST also views motor development as emerging from multiple systems, but structures this development within the specifics of known neural substrates based on neuroanatomical and neurophysiological evidence. Both dynamic systems theory and NGST open the door for multiple treatment options. Tenets of these theories would recommend that treatment for children with spastic CP include variable movement and sensory experiences to allow a child to explore the full range of sensorimotor options and select the most adaptable and functional motor patterns for a given stage of development. This type of treatment model may facilitate either a phase shift and change to a more functional attractor state, or facilitate selection of adaptive motor behavior.

Given the theoretical constructs of motor development in children with spastic CP, evidence for various treatment approaches for these children will now be reviewed. A brief overview of medical interventions and the impact they have on motor functioning in children with spastic CP will be followed by a more extensive consideration of behavioral treatments.

Medical Management for Children with Spastic CP

Medical management of motor signs in children with spastic CP includes pharmacological and surgical interventions. Pharmacological treatments for these children work to reduce increased tone and spasticity by either inhibiting excitatory neurotransmitters or increasing inhibitory neurotransmitters at the level of the spinal cord. Botulinum toxin (BOTOX) injections provide a treatment for addressing focal areas of spasticity or increased muscle tone. BOTOX works to block the release of the neurotransmitter acetylcholine from the presynaptic nerve terminal, which weakens the force of muscle contraction in hyperexcitable montoneurons. The use of BOTOX is most effective in conjunction with rehabilitative treatments, such as physical therapy (Goldstein, 2001). Surgical interventions are another option for treating children with spastic CP. Orthopedic surgeries are designed to facilitate maximal biomechanical advantages for limb function by carefully manipulating bones, tendons and joints (Goldstein, 2001). These surgeries can have positive and long lasting improvements for children with CP, while risks are relatively minimal. Selective dorsal rhizotomy is a treatment for lower limb spasticity and increased tone in children with CP. In this surgery, the sensory rootlets from the L2-S2 level of the spinal cord are cut. Changes following this procedure include an overall reduction in lower extremity tone and spasticity; however, success of this procedure is highly dependent on post-operative physical therapy to treat unmasked weakness in lower extremities (Brunstorm, 2001; Goldstein, 2001). Another surgical procedure is the implantation of an intrathecal Baclofen pump in the lumbar region of the spinal cord to diffuse Baclofen into the surrounding subarachnoid space. This method of Baclofen administration increases the concentration of the drug in the spinal region while minimizing the concentration in the brain (Goldstein, 2001).

Overall, medical treatments do a great deal to alleviate the positive motor signs associated with spastic CP; however, significant motor deficits can remain even when optimal medical treatment has been achieved. In recognizing the limitations of medical management and the potential for improvement in children with spastic CP, Brunstrom (2001) noted, "Although spasticity is one of the most tangible symptoms in patients with this group of heterogeneous disorders, these patients are more than just a composite of their spastic parts.... Most children with cerebral palsy are capable of doing far more and achieving at much higher levels than the medical profession or society would have them believe" (p. 14). Accordingly, behavioral treatments that target weakness, incoordination, sensorimotor integration, and endurance are required to maximize potential for functional improvement in children with spastic CP.

Behavioral Treatment for Children with Spastic CP

There are multiple behavioral treatment approaches that have been used for children with spastic CP over the years. One of the most well known and widely used is the neurodevelopmental treatment (NDT) from the work of Bobath and Bobath in the 1940s (Bobath, 1980; Bobath & Bobath, 1975). This approach is based on the neural maturationist theory of motor development. The goal of NDT is to minimize increased abnormal reflex patterns associated with hypertonus and facilitate more normal postural reactions with a progression towards normal functional movements (Bobath, 1980).

NDT therapists use handling and passive movement techniques to move children through controlled ranges of motion with a focus on the child's active reaction to the reflex inhibiting movements (Barry, 2001; Butler, 2001). Although NDT is widely used, a review of treatment efficacy literature for children with CP revealed that NDT is not currently supported by research based evidence (Butler, 2001). It was reported that there are few data to suggest this treatment model affects abnormal reflexes, muscle tone, or movement patterns nor has it been shown to have a positive effect on social-emotional development, cognitive development, or parent-child interaction (Butler & Darrah, 2001).

One of the more controversial aspects of treating children with spastic CP is strength training. The bias against strength training in these children suggests that many clinicians believe, "1) the increased physical effort associated with weight training would increase spasticity in muscles that were already over-active and thereby exacerbate joint stiffness and contractures; 2) that the children with CP would derive little or no benefit from weight training, since they lack sufficient isolated muscle control to increase strength in targeted muscles; and 3) that weakness, although present, is not considered a primary contributor to the motor dysfucntion" (Damiano, Vaughan, and Abel, 1995, p. 731). There is a growing body of literature rooted in muscle physiology and exercise science that contradicts these clinical objections and suggests strength training in children with CP is highly desirable (Damiano, & Abel, 1998; Damiano, Kelly, & Vaughan, 1995; Damiano, Martellotta, Sullivan, Granata, & Abel, 2000; Fowler, et al., 2001).

A number of studies have documented the positive benefits of strength training programs in children with CP (Damiano, & Abel, 1998; Damiano, Kelly, et al., 1995).

For example, Damiano, Vaughn, et al. (1995) reported that isolated muscle strength of the quadriceps improved in children with CP following a resistance training program, with no concomitant increase in hamstring strength (increased co-contraction). Similarly, Fowler et al. (2001) reported no evidence of increased spasticity following maximum effort exercises of quadriceps muscle in children with spastic CP. In sum, recent literature has not supported previous objections to strength training for children with CP.

Growing evidence from other non-conventional treatment concepts are also supporting a shift in methodology to more active and engaging treatment models (Bower & McLellan, 1992; Bower, McLellan, Arney, & Campbell,1996; Damiano, & Abel, 1998; Damiano, et al., 2000; Damiano, Kelly, et al., 1995; Damiano, Vaughn, et al., 1995; Fowler, et al., 2001; Gordon & Duff, 1999; Hadders-Algra et al., 1999; Schindl, Forstner, Kern, & Hesse, 2000; Valvano & Newell, 1998). This shift reflects changes in theories of motor development and the application of motor learning and skill acquisition principles to behavioral treatment in children with CP (Butler & Darrah, 2001). Concepts, such as intensive treatment, active and ample practice, and sensory augmentation are among those that are evolving in behavioral gait and limb treatment for children with CP.

Although the concept of intensive treatment may seem like an intuitive route to improve motor functioning in children with CP, it is rarely done. One reason why therapists may be hesitant to engage children with CP in intensive treatment is the fact that they tend to fatigue more quickly as compared to able-bodied children (Brunstrom, 2001). Brunstrom states, however, that "(i)t is not known how much the inherent muscle properties, movement difficulties, weakness, or hypertonicity in cerebral palsy contribute to a patient's energy inefficiencies or whether increased oxygen consumption is more directly related to the deconditioning effects of a sedentary lifestyle" (p. 13). Results from recent studies would suggest that the latter is true. Bower and colleagues documented that intensive treatment (daily, 1 hour sessions, 2-6 weeks) with specific treatment goals accelerated the acquisition of motor skills of children with CP (Bower et al., 1996, Bower & McLellan, 1992). In addition, parents, teachers, and caregivers reported a preference for the intensive mode of treatment as compared to a more traditional less intensive model (Bower & McLellan, 1992). Similarly, a study of intensive gait training in children with CP (25 minutes/day, 3 days/week, for 3 months) reported that eight of ten children gained significant benefit. In addition a majority of these children and families complied with, and reported enjoyment from, the intensive treatment model (Schindl, et al., 2000). These data support that at least some children with CP are capable of intensive treatment regimes, and fatigue may be due to de-conditioning effects, which would presumably decrease over the course of an intensive treatment program.

Incorporating active practice with repeated practice trials is supported theoretically by dynamic systems theory and NGST, as well as by motor learning and skill acquisition literature. Schmidt and Lee (1999) suggest that more learning will occur if there are more practice trials, all else being equal. Despite support for increased practice in treatment, the average number of repetitions of a task presented to a child with CP during physical therapy sessions has been reported to be less than five (Valvano &

Newell, 1998). Recent studies on task-specific, repetitive practice approaches (Schindl et al., 2000) and the use of active practice with increased practice trials (Gordon & Duff, 1999; Valvano & Newell, 1998) have been conducted in children with spastic CP. All studies have reported marked improvements in outcome measures including gait (Schindl et al., 2000), grip force production (Valvano & Newell, 1998) and anticipatory grip force precision (Gordon & Duff, 1999). Furthermore, to facilitate neuroplastic changes in the nervous system, repetitive practice must not be passive; rather, children must actively attend to movement and the accompanying sensory feedback (Byl, Merzenich, & Jenkins, 1996). Again, these approaches to treatment differ significantly from previous notions that emphasize tone-inhibiting maneuvers and gait –preparatory tasks, such as crawling and standing (Schindl et al., 2000).

Sensory deficits and difficulties with sensorimotor integration have been well-documented in children with CP (Evans, Harrison, & Stephens, 1991; Lesny, Stehlik, Tomasek, Tomankova, & Havlicek, 1993; Yekutiel, Jariwala, & Stretch, 1994). As a result, enhanced sensory input related to movement may be required for these children to learn new motor patterns. Gordon and Duff (1999) reported children with CP required additional blocked practice trials to develop anticipatory precision grip force for lifting objects of different weights and textures as compared to their typically developing peers. The authors suggested that children with CP may not be able to extract enough sensory information to form an internal representation of an object for anticipatory grip force from a single, or even a few practice trials. Hadders-Algra et al. (1999) demonstrated that explicit afferent information can facilitate task-specific modulation of postural adjustments during reaching of infants with CP. Weighted bracelets were placed on the arms of infants with CP, which improved the reach and facilitated appropriate postural adjustments that were not present during the unweighted condition. The authors suggested that deficits in sensory processing could be overcome, to some extent, by augmenting sensory information related to movement. Barlow, Finan, Bradford, and Andreatta (1993) have examined the benefit of enhanced sensory information from a dynamic actifyer (i.e., moving pacifier) on aberrant suck patterns of infants. They suggested that an enhanced oromotor sensory experience may facilitate selection of improved motor patterns for habilitation of the aberrant suck pattern generator.

Hadders-Algra summarized the importance of sensory augmentation in children with CP by stating, "Recent studies support the idea that deficits in sensory processing contribute to the variation in the scaling of motor output of children with mild to moderate forms of CP. The studies showed that practice, implying repetitions of self-generated sensory input and augmentation of movement related afferent information result in a decrease of variation in motor output and thus in a better task-specific adaptation of motor behavior" (Hadders-Algra, 2000b, p. 709). Therefore, treatments that enhance sensorimotor experiences and focus a child's attention on the sensations that accompany movement (intrinsic and extrinsic sensory feedback) may help establish improved movement patterns in children with spastic CP. In addition, the increased focus on sensory information may help children form an internal representation for a movement pattern (internal cue) which may improve carryover and maintenance of the new behavior (Fox, Morrison, Ramig, & Sapir, 2002; Ramig, Pawlas, & Countryman, 1995; Schmidt & Lee, 1999). Finally, the attention to sensory information associated with movement may be important for differentiation in cortical representations of motor movements (Byl et al., 1996).

In summary, recent literature examining behavioral treatment outcomes for gait and limb movement in children with CP have identified several important concepts including strength training, intensive treatment, active and ample practice and sensory augmentation and sensory awareness related to movement (Barlow, et al., 1993; Bower, et al., 1996; Bower & McLellan, 1992; Damiano, & Abel, 1998; Damiano, et al., 2000; Damiano, Kelly, et al., 1995; Damiano, Vaughn, et al., 1995; Fowler, et al., 2001; Gordon & Duff, 1999; Hadders-Algra et al., 1999; Schindl et al., 2000; Valvano & Newell, 1998). These themes are a departure from NDT, the most widely used treatment approach for children with CP; however, they are in accordance with recent theories of motor development and theories of motor learning and skill acquisition. Continued research in this area will clarify the role of these concepts in treatment for children with CP and further our understanding of motor development in this population.

Treatment for Speech

Although studies on motor development and treatment for gait and limb movement in children with CP have identified key concepts that may be important for successful behavioral treatment in this population (Barlow, et al., 1993; Bower, et al., 1996; Bower & McLellan, 1992; Damiano, & Abel, 1998; Damiano, et al., 2000; Damiano, Kelly, et al., 1995; Damiano, Vaughn, et al., 1995; Fowler, et al., 2001; Gordon & Duff, 1999; Hadders-Algra et al., 1999; Schindl et al., 2000; Valvano &

Newell, 1998), the application of similar concepts to treatment of speech disorders for these children has not been reported. Current speech treatment approaches for children with spastic CP include tone-inhibiting maneuvers associated with NDT, speech preparatory exercises associated with oro-motor training, prostheses for velopharyngeal insufficiency, and direct speech training associated with articulation therapy (Hardy, 1983; Solomon & Charron, 1998). There are limited reports on the efficacy of speech treatment for children with CP, hence, there is a great need for research in this area (Solomon & Charron, 1998; Yorkston, 1996).

The neuromotor bases of speech and voice disorders, which may guide treatment delivery models in children with spastic CP, are not well understood (Barlow & Abbs, 1984; Neilson & O'Dwyer, 1981; Workinger & Kent, 1991). Some proposed hypotheses of dysarthria associated with CP include "inefficient valving of the air stream caused by a generalized paresis of speech musculature; abnormalities of tone due to sustained background activity or spasticity of speech muscles; primitive or pathological reflexes interfering with articulatory control; imbalance between positive and negative oral responses caused by cortical lesions; and disruption of the voluntary control of speech muscles by random involuntary activity of the type associated with athetosis" (Neilson & O'Dwyer, 1981, p. 1013). Physiological studies in children with spastic CP to test these hypotheses are virtually non-existent; however, physiological based studies in adults with spastic CP may provide some insight.

To investigate the role of hypertonicity in disordered speech, Barlow and Abbs (1984) examined lip, jaw, and tongue isometric force production in six men with congenital spasticity. All subjects with spasticity were less stable in their ability to produce fine orofacial forces as compared to adults who did not have CP. The motor performance of men with spastic CP was more severely impaired in the tongue, a structure with relatively few muscle spindles, whereas, motor performance was less impaired in the jaw, a structure heavily innervated by muscle spindles. These observations did not support increased muscle spindle activity (spasticity) as being a primary cause of disordered speech in adults, and potentially children, with spastic CP.

Neilson and O'Dwyer (1981) examined EMG activity of lip, jaw, and tongue muscles in seven adults with CP (19-34 years of age) and normal control adults. They reported no evidence of weakness in individual articulator muscles as measured by integrated EMG; however, this did not preclude a functional weakness in the articulatory muscles caused by co-contraction of antagonist muscles. The authors reported consistent reproducibility of muscle pattern activation for subjects with CP. They suggested this was evidence of a consistent defect in motor programming. This defect may be the result of accurate motor commands being distorted by transmission through damaged descending pathways, inaccurate motor commands being formulated in the first place, or a combination of both. Taken together, limited physiological evidence suggests that speech treatment approaches for children with spastic CP may need to include tasks that facilitate both motor execution (strength training, sensory feedback, compensatory techniques) and motor learning (sensory feedback associated with movement, repeated practice trails, intensive training).

Recently, a model of speech treatment was developed that includes the concepts of strength/endurance training, repeated practice trials, intensity of treatment, and sensory awareness/augmentation. This treatment was developed for individuals with Parkinson disease and has been documented to be efficacious for that population (Ramig, Countryman, Thompson, & Horii, 1995). Changes following treatment in individuals with Parkinson disease, and select other adult neurological disorders, include improvements in vocal loudness, articulation, intonation, and rate (Dromey, Ramig, & Johnson, 1995; Ramig, Countryman et al., 1995; Ramig, Countryman, O'Brien, Hoehn, & Thompson,1996). This treatment (known as the Lee Silverman Voice Treatment, LSVT) has the essential concepts of: (a) exclusive focus on voice (specifically vocal effort and loudness), (b) stimulation of high effort productions with multiple repetitions, (c) delivery of treatment intensively (4 individual sessions a week for 4 weeks, 16 sessions in one month), (d) enhancing sensory awareness of increased vocal effort and loudness (calibration), and (e) quantification of behaviors (Ramig, Pawlas, et al., 1995). The specific techniques of LSVT bring together clinical concepts from literature in the areas of adult motor speech (Berry & Sanders, 1983; Duffy, 1995; Froeschels, Kastein, & Weiss, 1955; Hardy, 1967; Rosenbek & LaPointe, 1985; Wertz, 1975; Yorkston, Beukelman, & Bell, 1988) and voice (Aronson, 1990; Boone & McFarlane, 1988; Colton & Casper, 1996; Stemple, 1993). As such, LSVT offers a tool for examining the application of concepts that have been identified to be effective in limb and gait treatment of children with CP and motor speech functioning in adults with neurological disorders, on speech and voice functioning of children with spastic CP.

The rational for intensive treatment that includes ample practice and sensory augmentation/awareness has been defined. Accordingly, a brief rationale for focusing on voice in the treatment of motor speech disorders of children with spastic CP will be discussed. Children with spastic CP have disordered voice characteristics (Workinger & Kent, 1991), which may be due to muscle weakness, reduced central drive, incoordination of respiratory and laryngeal subsystems, or spasticity in speech musculature (Ansel & Kent, 1992; Keesee, 1976). Targeting increased vocal loudness may be desirable as it has been reported to improve both phonatory and articulatory stability in normal speech (Dromey & Ramig, 1998a) and in individuals with Parkinson disease (Kleinow, Smith & Ramig, 2001). In addition, training vocal loudness may serve as a "global" variable that affects multiple levels of speech production (respiratory, laryngeal, and orofacial) increasing motor output of them all (Dromey & Ramig, 1998a). Increased vocal effort and loudness has been associated with increased activity in the orofacial muscles (McHenry, 1997; Ramig, et al., 2000; Wohlert & Hammen, 2000) and may contribute to improvements in articulatory function. The relationship between increasing vocal effort and loudness and resonance has also been examined. Young, Zajac, Mayo, & Hooper (2001) suggested that increased loudness may facilitate velopharyngeal closure in a vowel-nasal context as measured by anticipatory nasal airflow. McHenry (1997) reported that estimates of VP orafice size were reduced as a function of increased loudness.

Another potential benefit of training increased loudness is that is does not involve deautomatization of speech production by requiring children to focus on specific speech parameters, such as rate, pauses, or articulatory precision; rather the child simple speaks louder (Dromey, 2000; Klienow et al., 2001; Sapir et al., in submission). This concept has been useful in treating individuals with Parkinson disease where the goal is rehabilitation of previously learned speech motor patterns (Fox, Morrison, Ramig, & Sapir, 2002). It is not known if the simplicity of the concept of speaking louder applies to children where the goal of treatment is habilitation of speech motor patterns (Scherzer, 2001). Similarly, the single instruction of speaking louder limits the cognitive demands associated with treatment, which may be important for children with low-average to below-average cognitive functioning. Finally, the target of increased vocal effort and loudness in treatment is elicited through modeling behavior (e.g., "do what I do") minimizing verbal instructions. Again, this limits cognitive load and allows the child's system to "self-organize" the best way to achieve the goal - consistent with concepts of dynamic systems theory.

Concerns related to using this type of approach with children with spastic CP include the potential for (a) vocal hyperfunction, (b) vocal or overall physical fatigue, and (c) increased associated movements or associated reactions with increased effort and loudness. In relation to vocal hyperfunction, the target of treatment is to train vocal loudness levels that are within normal limits for voice and speech output (i.e., not a shouted speech). In studies with individuals with Parkinson disease there was no evidence of vocal hyperfunction (ventricular hyperadduction or anterior-posterior foreshortening) post-treatment (Smith, Ramig, Dromey, Perez, & Samandari, 1995); rather, pre-treatment vocal hyperfunction in some individuals with Parkinson disease was reduced following treatment (Countryman, Hicks, Ramig, & Smith, 1997). Care and clinical skill in voice training prevents vocal hyperfunction in association with training increased loudness. Although fatigue has been a concern in children with spastic CP (Brunstrom, 2001), Schindl et al. (2000) reported that the initially "tiresome" training of an intensive gait treatment program was well tolerated by 8 of 10 children in their study. Similarly, children with spastic CP may initially experience fatigue with intensive speech treatment, which should diminish over time as the child builds strength and endurance (Schindl et al., 2000). Lastly, children with spastic CP are anticipated to have increased movement associated with stimulation of increased vocal effort and loudness. In contrast to NDT approaches that may use handling to inhibit associated movement, this approach will allow the increased movement to occur. The goal of treatment is to perturb the system (increase vocal effort and loudness), force it into a state of instability (explore extremes of sensorimotor experience related to increased vocal effort and loudness), and with repeated practice and intensive training, allow the system to "self-organize" into an improved motor pattern for voice and speech functioning (Kamm et al., 1990; Thelen, 1995).

In summary, children with spastic CP have disordered voice and speech characteristics, which limit functional communication and may negatively impact quality of life (Solomon & Charron, 1998; Yorkston et al., 1988). There are limited published outcome data on treatment approaches for these children (Yorkston, 1996). Recent developments in theories of motor development, behavioral gait and limb treatment, and speech treatment in individuals with Parkinson disease provide a solid framework (consistent with theories of motor learning) from which to test novel treatment concepts in children with spastic CP. Criticisms of previous behavioral treatment studies in children with CP, specifically NDT, have been that (a) treatment techniques were not delivered in a standardized manner, (b) there were not discrete dosages of treatment tested, and (c) that NDT techniques were often combined with other treatment approaches (Butler & Darrah, 2001). LSVT addresses these issues in that it has specific daily exercises prescribed for the entire treatment regime, there is a finite period of treatment, and the LSVT does not combine alternative treatment techniques (e.g. articulation or oromotor strategies).

The first purpose of this study is to examine the effects of an intensive long-term perturbation (behavioral treatment/LSVT) on speech and voice in children with CP. The specific questions related to this purpose are (a) Will intensive voice treatment (LSVT) perturb the speech motor system of children with spastic CP as evidenced by changes in acoustic measures of voice and speech? and (b) If speech is perturbed, will there be long-term changes in speech?

The second purpose of this study is to detect the presence of a therapeutic effect associated with this treatment, if there is one. Specific questions related to this purpose are (a) Do listeners prefer post-treatment speech samples over baseline speech samples of children with CP on select perceptual characteristics? and (b) Do parents perceive changes in perceptual characteristics related to their child's voice, speech, and communication skills after intensive speech treatment?

METHOD

Participants

Five children with a medical diagnosis of predominantly spastic cerebral palsy (CP) were recruited for this study. Children selected for the study were between 5 and 7 years of age and included both boys and girls. This age range was selected to target children of school age who may be highly motivated to communicate with peers (Scherzer, 2001). This relatively narrow age range also limited potential inter-participant variability due to physical size, general motor development, and linguistic development of the children included in the study. Additional selection criteria for the children with spastic CP included: (a) a perceptible speech or voice disorder that was characterized by disordered laryngeal and resonant characteristics, such as reduced loudness, loudness variability, monotone, breathiness, strain, or mild hypernasality, (b) hearing that was within normal limits, or aided to be within normal limits, (c) no vocal fold pathology (e.g. vocal nodules, vocal fold paralysis), (d) cognitive ability to follow directions and perform the tasks associated with the study protocol, and (e) stable medications. Children with a mild to moderate articulation disorder in addition to disordered laryngeal characteristics were eligible for inclusion; however, children with severe articulation disorders or severe velopharyngeal incompetence (e.g., need for palatal lift) were excluded. In addition, children with structural disorders of the speech mechanism (e.g. unrepaired cleft lip or palate) or a concomitant speech disorder, such as stuttering, were also excluded.

The medical diagnosis of spastic CP was confirmed by review of medical records. The characteristics and severity of speech, voice and oromotor functioning exhibited by participants with CP were determined by two certified speech-language pathologists from video samples of speech, voice, and oromotor tasks obtained during initial data collection sessions. The summary of limb and trunk involvement was determined by a certified physical therapist with extensive experience working with children with CP. Specific characteristics of each participant are described below.

A group of sex- and age-matched children were recruited to participate in this study to provide data against which data from the participants with CP could be compared. These children were between the ages of 5 to 7 years with no known neurological disease or condition and no history of speech or voice disorders.

Participant 1 (P1)

P1 was a 7 year, 10 month old boy with a diagnosis of spastic quadriparesis due to CP, and a secondary seizure disorder. His medications included Baclofen for spasticity and Tegratal for seizure control. These medications had been stable for the previous 3 years and remained stable from the pre-treatment through the post-treatment part of the study. Between post-treatment and follow-up sessions, P1 was being gradually taken off Tegratal. Overall body function for P1 was characterized by generalized weakness. He presented with weak trunk musculature and poor head control that was worse for midline (up-down) than side-to-side movements. He was unable to sit independently without support. P1 was wheelchair-dependent for mobility outside of the home (e.g. school) and used a commando crawl (prone position, pulling with elbows) to move around his house. He was able to support his full body weight and could assist on transfers.

Perceptual speech and voice characteristics included reduced loudness, monotone, intermittent hypernasality, imprecise consonants and a consistently slow rate of speech. The overall severity of his speech disorder was judged to be moderate. His oromotor function was characterized by slight asymmetry of facial movement, limited range of motion of the jaw and tongue, and a resting open mouth posture.

P1 was in an integrated classroom at school with daily pullout sessions for individual work and he had a full time aide to assist him. P1 had received speech therapy since he was 2 years old. At the time of the study, he was receiving speech therapy once a week with a focus on oromotor exercises. This speech treatment schedule did not change throughout the course of the study. His mother's concerns his for oral communication skills included his "breathy, quiet voice, and weak breath support," and that he did not have confidence when talking with other children. She reported that he could speak loudly when he was motivated (i.e., to get someone's attention in the home), but habitual speech was soft. She characterized his overall energy level as "low."

Participant 2 (P2)

P2 was a 5 year, 10 month old girl with a diagnosis of spastic quadriparesis due to CP. She did not take any medications. Her overall body function for P2 was characterized by generalized weakness, with relatively good isolated muscle control. She presented with mildly weak trunk musculature and good head control. She was an independent sitter. P2 demonstrated slight athetoid-like movements. These were characterized by movements that were initiated from the shoulder region and moved in large planes of direction; however, she had good isolated movements of hands and fingers. P2 could ambulate with assistance from a walker or a person. She used a wheelchair at school for mobility. She was able to pull to stand, support her full body weight and could assist on transfers.

Perceptual speech and voice characteristics included variable loudness, unnatural sounding prosody (e.g., equal emphasis), slow rate, and breathy, strained and sometimes whispered voice quality especially at end of sentences. She had mildly imprecise articulation that was mostly for words at the end of sentences. Her overall severity of speech disorder was judged to be moderate for respiratory and phonatory systems, and mild for articulation. Her oromotor function was only mildly affected and characterized by decreased range of motion for the tongue and jaw.

P2 was in a fully integrated classroom and she had an aide who assisted her with activities at school. Her parents reported that she had a history of speech therapy at various times in her life, but never consistently. According to her parents, goals of previous speech therapy included working on breath control with activities such as bubble blowing. She was not receiving speech treatment at the time of this study. Her parents' concerns regarding her oral communication skills included what they perceived as weak breath control, soft speech, and "choppy speech," which her parents described as "she took a lot of breaths to finish a sentence." Her parents reported that she was a very "chatty" girl who was willing to talk to anyone. They characterized her overall energy level as "medium."

<u>Participant 3 (P3)</u>

P3 was a 6 year, 1 month old boy with a diagnosis of spastic quadriparesis secondary to CP. He did not take any medications. Overall body functioning for P3 was characterized by increased tone and "stiffness" with minimal isolated muscle control. He presented with mildly weak trunk musculature and good head control. He was assymetrical in his degree of hypertonicity with the right side being more affected. P3 could sit independently with minimal support. He was wheelchair-dependent for mobility at school and home, but he could walk with assistance.

Perceptual speech and voice characteristics included variable loudness, strained voice quality with occasional voice stoppages, intermittent glottal fry, intermittent hypernasality, and inconsistent imprecise articulation. His overall severity of speech disorder was judged to be moderate. Oromotor functioning was characterized by asymmetry of facial muscles (left stronger than right), low tone in the face, an open mouth posture, and reduced range of motion in tongue and jaw movements. P3 received nutrition primarily though a G-tube, but he did take some food and drink orally.

At school, P3 was in an integrated classroom with a full-time aide. He had a history of consistent speech therapy since infancy. For the previous 1.5 years he had not had individual speech therapy outside of school. In school, he received speech therapy in a group twice a week. His mother's concerns for his oral communication included improving breath control and clarity of speech. She reported that he lost loudness and clarity at the end of sentences. She also reported he could "yell" when he wanted to get someone's attention or if he was mad. She described his overall energy level as "high."

Participant 4 (P4)

P4 was a 7 year, 7 month old boy with a diagnosis of spastic quadriparesis secondary to CP. He was taking Baclofen for spasticity, which was kept stable throughout the course of the study. His overall body involvement was characterized by significant weakness, with increased tone present in upper > lower extremities. He presented with very weak trunk musculature and poor head control in all directions. He was not an independent sitter. P4 used some commando crawling and rolling for mobility at home and was wheelchair-dependent for mobility outside of the home.

Perceptual speech and voice characteristics included reduced loudness, monotone, breathy voice, and imprecise articulation. P4 had a paucity of speech output that was limited to 1 and 2 word phrases, which was often produced with whispered speech. His overall speech and voice disorder was judged to be moderate to severe. Oromotor functioning was characterized by asymmetry of facial musculature (left stronger than right), drooling, a tongue thrust, and decreased range of motion of the jaw and tongue. P4 received nutrition from a G-tube and took minimal food or drink orally.

In the school setting, P4 was in a special education classroom for at least half of the day and in an integrated classroom the remainder of the day. P4 had a history of previous speech therapy that focused on breath control for speech and use of augmentative communication devices. He was not receiving any speech therapy during this study. His mother's concerns for his oral communication included his limited communicative abilities (decreased language skills and whispered speech output), breath control, and his quiet, "whispered" voice. She wanted him to be able to speak in longer utterances (greater than 1-2 words) that could be heard and understood. She characterized his overall energy level as "high".

Participant 5 (P5)

P5 was a 6 year, 7 month old girl with a diagnosis of spastic quadriparesis secondary to CP. She did not take any medications. Her overall body involvement was characterized by generalized weakness, relatively normal tone, with spasticity. She presented with mildly weak trunk musculature and good head control. She could sit independently. She was able to ambulate with assistance from a walker or a person, but was primarily wheelchair-dependent for mobility at home and school.

Perceptual speech and voice characteristics included variable loudness, mildly monotone, and a strained voice quality especially at the end of sentences. Her overall speech disorder was characterized as mild. Oromotor function was mildly involved. There was a slight decrease in jaw range of motion and speech was perceived to be effortful as evidenced by occasional eye-squinting and other facial grimacing during speech (as if in an effort to "squeeze" out speech).

P5 was in a fully integrated classroom with a part-time aide who assisted her with activities at school. She had no history of speech therapy. Her parents' concerns for her oral communication included not being able to "take a full tidal breath," not always having enough breath, stopping in the middle of a sentence to get a breath, and breathing coordination. Her mother characterized her overall energy level as "medium."

Design

The purpose of this study was to examine the effects of an intensive treatment on speech and voice functioning in children with CP. Thus, a study design appropriate to treatment outcome research was used. The initial goal of treatment outcome research is to simply detect any therapeutic activity, as well as any negative side effects, associated with treatment (Robey & Schultz, 1998). Methods used during this initial Phase I research include case study designs, small group experiments, and single subject designs. During Phase I research there is only one active treatment in the design, sample sizes are small, there is a lack of strict external controls, and Type I and Type II errors are tolerated (Robey & Schultz, 1998).

For this study, a multiple baseline design with replication across participants (Barlow & Hersen, 1984; McReynolds & Kearns, 1983) was used. This design is preferred over a withdrawal or reversal design (e.g., A-B-A-B) because the targeted behavior in this study (increased vocal effort and loudness) was not expected to return to baseline levels after the initiation of treatment (Ingham & Riley, 1998; McReynolds & Kearns, 1983). In a multiple baseline design with replication across participants, the treatment variable (intensive treatment) was applied sequentially to the same behavior (vocal output) across different, but matched participants sharing the same environmental conditions (Barlow & Hersen, 1984).

Ideally, all participants in this type of design participate in the study concurrently. Every attempt was made to study the present participants concurrently; nevertheless, this was not possible due to logistical challenges and scheduling conflicts with participants and families. Therefore, a non-concurrent multiple baseline design was used, which weakens the strength of the design (Barlow & Hersen, 1984). In this design, duration of baseline periods and time of treatment initiation were predetermined for each replication in the study. Participants were then randomly assigned to one of the predetermined study conditions.

Procedures

Participant Selection

Potential participants were screened to determine if they met study criteria. Upon initial contact with the parents or legal guardians of a potential participant, a telephone screening questionnaire was completed (Appendix A). This telephone interview confirmed the age, sex and medical diagnosis of a child. In addition, information was gathered regarding the parent's perceptions of their child's speech and voice difficulties. The study protocol was explained to parents and all questions were answered. If parents were interested in having their child participate in the study, a screening session for the child was scheduled. The screening session included: (a) a brief voice and speech screening to confirm that the speech disorder was primarily laryngeal and resonant in nature, (b) screening of cognitive ability to see if the child could follow directions and perform the basic tasks of the study protocol (sustain a vowel, repeat words/phrases), and (c) a hearing screening (at 500, 1000, 2000, 4000 at 25 dB HL). In addition, a laryngeal examination of each child was conducted by an otolaryngologist to ensure that no laryngeal pathology existed that would preclude a child's participation in this study.

Study Conditions

Participants in this study were randomly assigned to one of four study conditions. Each condition consisted of recording sessions and treatment sessions. The conditions were as follows:

Condition A: 16 treatment sessions (4 sessions a week for 4 consecutive weeks); up to 4 recording sessions during the 2 weeks prior to the start of treatment (baseline); 2 recording sessions 1 week immediately following treatment (post-treatment); 2 recording sessions 6 weeks after the conclusion of treatment (follow-up).

Condition B: 16 treatment sessions (4 sessions a week for 4 consecutive weeks); up to 6 recording sessions during the 3 weeks prior to the start of treatment; 2 recording sessions 1 week immediately following treatment; 2 recording sessions 6 weeks after the conclusion of treatment.

Condition C: 16 treatment sessions (4 sessions a week for 4 consecutive weeks); up to 8 recording sessions during the 4 weeks prior to the start of treatment; 2 recording sessions 1 week immediately following treatment; 2 recording sessions 6 weeks after the conclusion of treatment.

Condition D: up to 4 recording sessions during the course of 2 weeks; 2 recording sessions 4 weeks after the initial recording sessions; 2 recording sessions 10 weeks after the initial recording sessions. For the participant in condition D, 16 treatment sessions (4 sessions a week for 4 consecutive weeks) were offered following the final recording session.

Participant assignment to study conditions and the actual number of recording sessions in the baseline, post-treatment, and follow-up phases are summarized in Table 1.

Recording Sessions

Equipment and Set-up

Data collection procedures were identical for all recording sessions (baseline, post-treatment, and follow-up). Duration of each session ranged from 30 minutes to 1 hour. All data were collected in an IAC sound-treated booth. Four of the participants were comfortably seated in their own wheelchair. One participant sat in an adaptive chair, which provided postural support. The seating arrangement was the same for all data collection sessions.

Audio recordings were made with the participant wearing a small omni-directional condenser microphone (Audio-technica, Model AT 803b) taped to the forehead and secured with a soft, elastic cloth headband. This method allowed for mouth-to-microphone distance to be kept at a constant 4 inches throughout the recording session. Microphone signals were recorded onto a digital audiotape (DAT) recorder (Panasonic Digital Audio Tape Deck, Model SV-3500). A calibration signal (960.5 Hertz) was recorded prior to the start of the session and again at the end of the session. Standard procedures for recording calibration signals were followed. Specifically, the omni-directional condenser microphone was taped to a Styrofoam head at a distance of 4 inches from the mouth (on the Styrofoam head). A sound level meter microphone was placed next to the condenser microphone at the same distance from the mouth (4 inches). A tone generator (Korg Auto Chromatic Tuner, AT-12) was placed in the same plane as the Styrofoam head's mouth. The generated tone was recorded onto the DAT tape and the exact sound pressure level (SPL) reading in decibels was noted.

Interviews

The participant with CP was interviewed during the initial baseline session, the initial post-treatment session, and the initial follow-up session (See Appendix B). The participant was asked questions about his/her speech to find out what he/she did or did not like about talking and how he/she felt about his/her ability to talk with other children and adults. In addition a brief oral motor examination was completed (Darley, Aronson, & Brown, 1975). During one of the baseline sessions the participant's parent was interviewed and asked similar questions about his or her child's speech. The parent was also asked about the participant's daily activities (e.g. additional therapies, extracurricular activities) and social interactions outside of the school environment. During one of the post-treatment sessions, the participant's parent was interviewed and asked questions about any changes they had observed in their child's speech following speech treatment, and about their general impressions of the treatment approach used in this study (Appendix C).

Speech and Voice Tasks

During recording sessions, participants were asked to perform a variety of speech and voice tasks including: (a) producing maximum duration sustained vowels (e.g., "take a deep breath, and say 'ah' for as long as you can," (b) producing maximum fundamental frequency range (e.g., "go as high as you can on 'ah'" "go as low as you can on 'ah'"), (c) repeating a series of short sentences, and (d) watching a short cartoon video then retell what happened (McNeil, 1992) (See Appendix D for protocol). The order of presentation of these tasks was randomized across all recording sessions. The task of retelling a cartoon was elicited once during the baseline, post-treatment, and follow-up recording sessions (rather than during all sessions). All recording sessions were videotaped.

The maximum performance voice tasks (sustained vowels, frequency range) were designed to examine the upper limits of performance for a participant on a given voice task (Kent, Kent, & Rosenbek, 1987). Multiple repetitions of each task (4 to 15 trials) were collected at all recording session (Kent et al., 1987). It has been documented that children with spastic CP perform more poorly on these types of tasks than their typically developing peers (Wit, Maassen, Gabreels, & Thoonen, 1993). The data collectors who elicited maximum performance tasks were careful to use specific instructions that encouraged maximum performance from participants.

Speaking tasks were also included (Kent & Kent, 2000; Netsell, 1983). The participants repeated a series of short sentences three times each recording session ("Buy Bobby a puppy," "The potato stew is in the pot," "The blue spot is on the key"). When sentence repetition was used, the data collectors were careful to model the sentences without exaggerated loudness, articulation, or pitch inflection. Spontaneous speech was elicited by having participants watch a short cartoon video (*Canary Row*, Warner Brothers) then retell what happened to a listener (McNeil, 1992). Participants were instructed to retell the events of the cartoon to a parent or sibling who had not seen the video. The parent or sibling was encouraged to ask the participant questions about what he/she saw in the cartoon, or ask questions for clarification.

Perceptual Rating Forms

Perceptual rating scales were filled out by the participant's parent at baseline, post-treatment, and follow-up recording sessions. A visual analog scale (Kempster, 1984; Schiffman, Reynolds, & Young, 1981) was used to obtain ratings on nine variables related to voice (e.g. loudness, nasality, hoarseness, monotone, strain), speech (understood by others), and spoken communication (participate in conversation, start conversation, frustration with communicating) (Appendix E). The scale required the parent to place a slash through a solid horizontal line that represented a continuum ranging from continuous presence of a characteristic, (e.g., "Voice is always loud enough") to complete absence of a characteristic, (e.g., "Voice is never loud enough"). Written and verbal instructions were provided for the perceptual form and all questions related to the form were answered. A clean unmarked visual analog form was provided for the parent each time he or she filled one out (without reference to ratings from previous recording sessions).

At the initial post-treatment recording session, the child's parent filled out a "Treatment Rating Form" (Appendix F). This form included 14 questions related to the parent's impression of the participant's treatment outcome and general impressions of the treatment approach. Parents responded to questions, such as "How much do you think that your child's recent voice therapy <u>improved</u> your child's voice overall?" on a 5 point rating scale (1 – Not at all, 2 – Very little, 3 – Somewhat, 4 – Quite a bit, 5 – A lot).

<u>Data Collectors</u>

Data were collected by individuals (a pool of 4) who were well-trained in the experimental protocol. The same group of individuals collected all the voice recording session data (baseline, post-treatment, and follow-up sessions). In addition, these data collectors conducted all participant and parent interviews. The investigator who delivered treatment did not collect data in recording sessions for participants with CP.

Intensive Voice Treatment

The treatment model used in this study was LSVT (Ramig, Pawlas, et al., 1995). Modifications to the concepts of LSVT or treatment tasks were not made; however, therapy activities were adapted to be appropriate for children with significant motor impairments. Treatment consisted of 16 sessions (1 hour each) of voice treatment delivered 4 days a week for 4 weeks. All treatment sessions were conducted in the participant's home. The focus of treatment was on increasing vocal effort and loudness for speech. In LSVT, the cue of "loud" is frequently used to elicit increased vocal effort and loudness. In this study, the cues "strong voice" or "big voice" were used in addition to the cue "loud voice" to elicit these behaviors.

The first half of each treatment session consisted of "daily tasks" (Ramig, Pawlas, et al., 1995). These tasks included: (a) maximum duration sustained vowels (e.g., "take a deep breath and say 'ah' as long as you can using your strong voice"), (b) maximum frequency range ("go as high as you can on 'ah'/go as low as you can on 'ah' with your strong voice"), and (c) repeating 10 functional phrases (generated by the participant and his/her family) five times each ("say these phrases with your strong voice that you used during the 'ah' exercise"). One purpose of these exercises was to facilitate respiratory and laryngeal coordination for voicing, while building strength and endurance of the voice. A second purpose of these exercises was to use the voice tasks (long "ahs," high/low "ahs") to shape good voice quality. For example, if the child had a strained, pressed or breathy voice quality, the investigator would use various shaping techniques, such as modeling (e.g., "Do what I do"), to improve voice quality. The second half treatment sessions was spent on training the vocal loudness and quality achieved during daily tasks into speech. The training progressed through a hierarchy of difficulty beginning with single words and working up to conversational speech whenever possible.

All exercises included repeated practice trials (i.e., minimum of 15 repetitions of each training task), while incorporating sensory augmentation (cueing increased vocal effort and loudness) and sensory awareness (e.g., "Did you feel your voice? Did you hear how you sounded?"). Homework exercises, which included treatment tasks (e.g., long "ahs," high/low "ahs," and speech exercises using the participant's loud/strong/big voice), were assigned on a daily basis. In addition, carry-over exercises, such as "Say good morning to the bus driver using your 'STRONG VOICE'" or having the participant leave voice mail messages to the investigator who delivered treatment, were given.

The participants displayed a variety of spontaneous movements accompanying increased vocal effort during voice and speech tasks. These movements, which were perceived as natural age-appropriate communicative gestures associated with speech (even though these movements were distorted due to motor signs in limbs), were accepted and not modified in any manner. This is in contrast to previous speech treatment approaches that may have altered or limited movement accompanying speech of children with CP to patterns that appeared "normal" (Hardy, 1983; Solomon & Charron, 1998). Care was taken to watch for associated reactions or associated movement (e.g., throwing back the head, squinting the eyes) with stimulation of increased vocal effort that would not be desirable for the participant's functional communication goals, and if they occurred, movement patterns were modified through modeling behavior (e.g., "say 'ah' like I do").

Movement in participants with CP was also paired with speech to facilitate maximum sensory augmentation and sensory awareness experiences. For example, increased duration of sustained vowels was facilitated by moving the participant's arms or by bouncing on a ball with the investigator. Physical activities, such as throwing balls, punching bags, rolling on the floor, were used in conjunction with speech and voice tasks to arouse and stimulate the participant. Variations in posture were explored such as sitting upright with varying levels of support, lying on the stomach, or lying on the back. Occasionally, the investigator touched the participant's throat or placed her hands on the rib cage of the participant to draw sensory attention to these areas with the instructions to "say 'ah' as long as you can with your loud/strong/big voice."

A tape recorder was used during treatment sessions to provide the participant with auditory feedback regarding his/her voice and speech production. Periodically, treatment sessions were videotaped.

Participation of Typically Developing Peers

The typically developing peers participated in two recording sessions, completing the same tasks as the participants with CP (see Table 1). These children did not participate in the laryngeal examination or the treatment phase of this study.

Data Analysis

Data Preparation and Digitizing Procedures

All acoustic data were reviewed and problematic samples were excluded from further analysis. Problems included clipped signals (approximately 10-15 samples across all participants and sessions) and faulty microphone signals (one recording session for two participants). Samples from the DAT tapes (including the calibration tone) were digitized at 22.05 kHz with the acoustic analysis program *Praat* (Boersma & Weenick, 2002) installed on a Gateway 2000 computer. Each sample (e.g. sustained vowel, sentence) was digitized and saved in an individual wave file for further analysis. The calibration tone for each recording session was analyzed for decibels of sound pressure level (dB SPL) using the *Praat* program. The dB SPL value from the *Praat* program was subtracted from the dB SPL value recorded from the SLM during calibration procedures to create a "correction factor." The correction factor was added to all dB SPL values produced by the *Praat* program across voice and speech tasks.

Acoustic Analysis

Analysis of Voice Tasks

For each sustained vowel sample (longest, highest, lowest), the *Praat* program was used to display the acoustic waveform, the intensity contour (dB SPL) and the fundamental frequency (f_0) contour in Hertz (Hz). From the intensity contour, the following measures were obtained: (a) duration, and (b) mean (and SD) dB SPL. The acoustic waveform and pitch contour were then inspected for any breaks or variations in voicing that interfered with the pitch analysis, such as glottal fry. Any segments of glottal fry were cut from the acoustic waveform. The pitch contour was then extracted from the remaining acoustic waveform. From the pitch contour the following measures were obtained: (a) minimum f_0, (b) maximum f_0, (c) mean (and SD) f_0. These values were recorded in an excel spreadsheet for further analysis.

Maximum performance tasks.

For maximum sustained vowel durations, the single longest vowel duration per recording session was identified and graphed for visual analysis. For maximum f_0 range, the single highest and single lowest vowel frequency value produced per recording session were determined and graphed for visual analysis. The maximum f_0 range was calculated (maximum frequency – minimum frequency) and displayed visually. Grand means (and SDs) and mean differences were calculated for baseline sessions (Base), post-treatment sessions (Post), and follow-up sessions (FUP) for these measures.

Sustained phonation.

The three longest duration vowels were chosen for sustained phonation analysis (with the exception of one participant, one session with only two). The following measures were obtained for each of the 3 longest vowels per recording session, (a) mean (and SD) dB SPL, (b) mean (and SD) f_0, and (c) harmonics to noise ratio (HNR). For HNR measures, the middle 30-second segment of each vowel (selected by determining the midpoint duration of the vowel and taking 15 seconds prior to and immediately following the midpoint) was used in the analysis. HNR was included as an acoustic measure of long-term phonatory stability that may be related to the perceptual characteristic of voice quality (Hartelius, Buder, & Strand, 1997; Ramig, Scherer, Klasner, Titze, & Horii, 1990). The mean (and SD) value of the three longest vowels for each of these measures per recording session were determined and displayed for visual analysis. Grand means (and SDs) and mean differences were calculated for Base, Post, and FUP on these measures

Analysis of Speech Tasks

Each digitized speech sample from the sentence repetition and cartoon description tasks were filtered through a customized analysis program (Story, Matlab, 2002) that removed pauses and voiceless consonants. This program created a new file for each speech sample that contained only voiced segments. These new files were analyzed in the same manner as the vowel samples described above. For each speech sample, the following values were obtained: (a) mean (and SD) dB SPL, (b) minimum f_0, (c) maximum f_0, and (d) mean (and SD) f_0. These values were recorded in an excel spreadsheet for further analysis.

For each recording session, the (a) mean (and SD) dB SPL, (b) mean (and SD) f_0, and (c) f_0 range of nine sentence repetitions were calculated and graphed for visual analysis. Grand means (and SDs) were calculated for Base, Post, and FUP on these measures. Similarly, for each recording session where the cartoon description task was elicited, the above measures were calculated for each sentence. The total number of sentences produced by a participant varied across sessions and participants ranging from as few as 10 to as many as 30. Grand means (and SDs) and mean differences were calculated for Base, Post, and FUP on these measures.

Auditory-Perceptual Analysis (Listener Task)

A pair-comparison listening task with a group of seven experienced listeners was conducted. The listeners were all certified speech-language pathologists who had extensive experience across the areas of motor speech disorders, voice, and children with CP. The sentence repetition task from the final Base session, the final Post session and the final FUP session were used. Each speech sample included three repetitions of a sentence ("Buy Bobby a puppy," "The blue spot is on the key," "The potato stew is in the pot") produced within a given session. The three different sentences were paired as follows: (a) Base "Bobby" to Post "Bobby," (b) Base "Bobby" to FUP "Bobby," (c) Post "Bobby" to FUP "Bobby," (d) Base "Key" to Post "Key," (e) Base "Key" to FUP "Key," (f) Post "Key" to FUP "Key," (g) Base "Potato" to Post "Potato," (h) Base "Potato" to FUP "Potato," and (i) Post "Potato" to FUP "Potato." To measure listener reliability, three of these pairs were randomly selected and repeated for each participant. The total number of pair presentations per participant was 12, with the exception of P2 who only had 10 pairs (due to her unwillingness to repeat the sentence "Potato" at Post). The order of presentation of sentences within each pair (e.g. Base to Post, Base to FUP, Post to FUP) and the order of presentation of the12 pairs were randomized for each participant.

Listeners were seated comfortably in a chair at a desk with a computer and speakers on it. Listeners were asked to listen to a pair of speech samples (each pair containing three repetitions of a sentence) and to rate which sample they "preferred" for the following variables: overall loudness, loudness variability, overall pitch, and pitch variability. Listeners were asked to mark on a rating form which sample they "preferred", Sample A, Sample B, or No Preference. The listeners were asked to provide comments about why they chose one speech sample over another (see rating form, Appendix G). After rating loudness and pitch, the listeners were asked to listen to the paired speech samples again and rate which they "preferred" for overall voice quality and articulatory precision. Listeners were given the option to provide comments about why they chose one speech sample over another.

Listeners were allowed to adjust the volume control on the speakers to a loudness level that was comfortable for each participant. Once the volume control was set for a given participant, it did not change at anytime during the listening task for that participant. Listeners were allowed to listen to the speech samples as many times as they wanted prior to making their decision. Listeners were allowed to take breaks at anytime. The listening task took approximately 2 hours to complete.

For each of the six perceptual variables rated (overall loudness, loudness variability, overall pitch, pitch variability, overall voice quality, articulatory precision) a total of 63 choices for preference (9 pairs X 7 listeners) were made per participant (49 choices for P2). The number of times a speech sample from the Base session, Post session, FUP session was chosen, or a choice of "no preference" was made, were tabulated. Percentage scores for preference of speech samples were calculated by dividing the frequency of preference of a given category by the total number of choices for that perceptual variable (e.g. 10 choices for Base samples/63 total possible choices = 16% preference for Base speech samples). These percentage scores for all six perceptual variables were graphed for visual analysis.

The three repeated pairs per participant were analyzed for agreements in choice of preference from the first presentation of a pair to the repeated presentation. Agreement ratings were marked and summed. A percentage of within listener agreement was calculated from the summed values, and reported as an index of within-listener reliability. Across the seven listeners, within-listener reliability ranged from 74 – 89%.

Analysis of Parent Perceptual Ratings

Standard procedure for analysis of visual analog scales was used to examine perceptual data (Boeckstyns & Backer, 1989). The total distance of the line representing the continuum of presence or absence of a characteristic was measured. The distance of the slash on the line from one end of the continuum was measured and calculated into a percentage based on the total distance of the line. This percentage represents the parents' perceived presence of a particular characteristic in the child with CP "most of the time." Analysis of the "Treatment Rating Form" involved tabulating the parents' rating responses (1 – Not at all, 2 – Very little, 3 – Somewhat, 4 – Quite a bit, 5 – A lot) to each of the 14 questions. Comments from the parents' post-treatment interview were transcribed from videotapes of the interviews.

Determination of Treatment Effects

Evaluation of treatment effects in single subject designs is based primarily on visual inspection of data (Barlow & Hersen, 1984; Ingham & Riley, 1998; McReynolds & Kearns, 1983). Visual inspection involves identifying trends (change of direction), slope (steepness of change in direction), and level (frequency of occurrence of behavior) in data (Barlow & Hersen, 1984; McReynolds & Kearns, 1983). Trend, slope and level observations can be used to examine stability of baseline performance, overlap between scores of adjacent phases, changes in trend between phases, and changes in level between phases. There are no well-defined standards for evaluating treatment effects from trend, slope, and level analysis in single subject designs (McReynolds & Kearns, 1983). In general, changes across phases must be large enough to allow no room for ambiguous interpretation during visual inspection.

In this study, participant data for acoustic measures were displayed graphically for visual analysis. This manner of presentation allows for both visual inspection of trends within participants as well as patterns of change across participants (Barlow & Hersen, 1984; McReynolds & Kearns, 1983). Visual analysis of data graphs were used to: (a) examine stability of baseline performance, (b) observe trends within study phases, and (c) evaluate trends across study phases. It was recognized that baseline data showing trends (instability) were less predictable for determining treatment effects as compared to stable baseline performances (McReynolds & Kearns, 1983). When visual trends in data were observed, the individual values for a measure within a phase (e.g., Base) were examined for overlapping values between study phases (Base to Post). Visually observed trends that were accompanied by non-overlapping values between study phases were considered "strong" trends and indicative of a treatment effect. Visually observed trends there were accompanied by overlapping values (e.g., due to variable Base performance) were noted, but not considered indicative of a treatment effect.

Participant data for listener preference ratings and parent perceptual ratings were also displayed graphically. Changes in level (e.g., frequency of preference expressed as a percentage) between study phases (Base, Post, FUP) were visually examined for trends (Barlow & Hersen, 1984; McReynolds & Kearns, 1983). Visually observed trends that were accompanied by a change in level of 20% or more were considered "strong" trends and indicative of a treatment effect.

RESULTS

Data for each participant with CP are presented individually. Comparisons to data from typically developing peers are included (Appendix H). Patterns of behavior across participants are summarized at the end of this section.

Data for P1

Maximum Performance Tasks

Maximum performance data for P1 are displayed in the first graph of Figures 2 and 3. Grand means (and SDs) and mean differences for these measures are presented in Table 2. Visual inspection of maximum duration of vowel data revealed a strong increasing trend (i.e., consisting of non-overlapping values between study phases) for duration from Base to FUP. For maximum highest frequency, a strong increasing trend was observed from Base to Post, and Base to FUP. For minimum frequency, a strong decreasing trend was observed from Base to Post. For maximum frequency range data there was a strong increasing trend observed from Base to Post, and Base to FUP.

Sustained Phonation (Three Longest Vowels)

Mean vocal SPL, f_0, and HNR data of the three longest sustained vowels for P1 are displayed in the first graph of Figures 4, 5, and 6, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 2. Visual inspection of vocal SPL data revealed a variable performance across all study phases with no strong trends between phases. Visual inspection of f_0 and HNR data for sustained vowels revealed no strong trends across study phases for these measures.

Sentence Repetition

Vocal SPL, f_0, and f_0 range data for P1 during sentence repetition are displayed in the first graph of Figures 7, 8, and 9, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 2. Visual inspection of vocal SPL and f_0 data revealed no strong trends across study phases. For f_0 range data, a strong increasing trend from Base to Post (non-overlapping values) was observed.

Cartoon Description

Means (and SDs) and mean differences for vocal SPL, f_0, and f_0 range across study phases are presented in Table 2. Trends in data are not discussed for the cartoon description task due to a single data point per acoustic measure at Base, Post, and FUP. Vocal SPL did not change from Base to Post, increased from Base to FUP (2.6 dB SPL). Mean f_0 decreased slightly from Base to Post, and then increased slightly from Base to FUP. F_0 range values did not differ from Base to Post, but increased from Base to FUP by 38 Hz.

Listener Ratings

Percentage of listener ratings for preference of speech samples spoken at Base, Post, and FUP are presented in the first graph of Figures 10-15. Visual inspection of data revealed strong trends (i.e., accompanied by a 20% or more increase) for preference of Post speech samples as compared to Base. Articulatory precision was the perceptual variable that listeners had the greatest preference for Post speech samples over Base samples. There was a trend (not strong) for listener preference of Base speech samples over FUP across all perceptual variables. These data indicate that immediately Post, P1 demonstrated perceptual characteristics in speech that the listeners preferred over Base speech samples. However, by FUP, preference data suggested that perceptual characteristics of P1's speech returned to Base levels, or worse.

Parent Perceptual Ratings

Parent perceptual ratings are displayed in the first graph of Figure 16. Visual inspection of the data identified several strong trends (i.e., accompanied by a 20% or more change) across Base, Post, and FUP ratings. Improved perceptions of loudness, hoarseness, breathiness, initiation of talking, and frustration with talking were observed from Base to Post, and participation in talking increased from Base to FUP. Parent ratings of improved perceptions of hoarseness and initiation of talking were maintained from Base to FUP, while ratings for loudness, breathiness, and frustration with talking were not.

Parent responses to the 14 questions on the treatment rating form were all rated a 4 (quite a bit) or 5 (a lot). The exception was question 5 ("To what extent do you think therapy made your child's voice clearer?") which was marked a 3 (somewhat).

Summary of Results for P1

Across all maximum performance tasks, P1 demonstrated improved values for acoustic measures from Base to Post, which were maintained above Base values at FUP. For sustained phonation measures, no strong trends were observed across study phases; however, an average increase in HNR values from Base to FUP (2.0 decibels) was noted. For speech tasks, there was a strong increasing trend for range between Base and Post in sentence repetition and notable improvements across all measures from Base to FUP for cartoon description. Changes in acoustic measures for P1 that moved him in the direction of performance of his typically developing peers included all values for maximum performance tasks, HNR values for sustained phonation, and vocal SPL and f_0 range values for sentence repetition and cartoon description (Base to FUP only for cartoon). For all other acoustic measures, there was either no change in performance or a change that moved P1 away from the performance range of his typically developing peers. Listener ratings revealed a strong preference for perceptual characteristics of Post speech samples over Base, and Base speech samples over FUP. Overall parent impressions of P1's response to treatment and impressions of the treatment technique at Post were favorable, with maintenance of a few improved perceptions at FUP.

Data for P2

Maximum Performance Tasks

Maximum performance task data for P2 are displayed in the second graph of Figures 2 and 3. Grand means (and SDs) and mean differences for these measures are presented in Table 3.

Visual inspection of maximum duration of sustained vowel data revealed a strong increasing trend for duration from Base and Post, and Base and FUP. In the second graph of Figure 2 there are missing data points for maximum and minimum f_0 on Base1 and Base 3, and for minimum f_0 on FUP 1, due to P2's refusal to perform the tasks on these days. Visual inspection of data for maximum highest f_0 revealed an increasing trend from Base to Post and a strong increasing trend from Base to FUP. Visual inspection of minimum f_0 data revealed a decreasing trend from Base to FUP (i.e., non-overlapping values). Maximum f_0 range data revealed a strong increasing trend from Base to Post, and Base to FUP. These data indicate that although P2's Base performance varied, maximum f_0 range increased beyond the level of Base instability, as evidenced by non-overlapping values between these phases.

<u>Sustained Phonation (Three Longest Vowels)</u>

Vocal SPL, f_0, and HNR data of the three longest sustained vowels for P2 are displayed in the second graph of Figures 4, 5, and 6, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 3. Visual inspection of vocal SPL and f_0 data revealed a strong increasing trend from Base to Post and Base to FUP. Visual inspection of HNR data revealed no strong trends across study phases.

<u>Sentence Repetition</u>

Vocal SPL, f_0, and f_0 range data for P2 are displayed in the second graph of Figures 7, 8, and 9, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 3. Visual inspection of vocal SPL and f_0 data for sentence repetition revealed strong increasing trends from Base to Post, and Base to FUP. Visual inspection of f_0 range data revealed a strong increasing trend from Base to FUP.

<u>Cartoon Description</u>

Means (and SDs) and mean differences on measures of vocal SPL, f_0, and f_0 range for P2 are presented in Table 3. There was a marked increase in vocal SPL from Base to Post (13.2 dB SPL) and Base to FUP (5.8 dB SPL). Mean f_0 also markedly increased from Base to Post (166 Hz) and Base to FUP (62 Hz). Finally, a large increase in f_0 range was observed across study phases.

Listener Ratings

Percentage of listener ratings for preference of speech samples spoken at Base, Post, and FUP are displayed in the second graph of Figures 10-15. Visual inspection of the data identified strong trends for preference of Post speech samples over Base for overall loudness, loudness variability, pitch variability, and overall voice quality. Overall loudness was the perceptual variable for which listeners had the greatest preference for Post speech samples over Base. Strong trends for preference of FUP samples over Base were identified for overall loudness, pitch variability, overall voice quality and articulatory precision. There were no strong trends observed between percentage scores for preferences between Post or FUP speech samples. These data indicate that changes in perceptual speech and voice characteristics of P2 that influenced listeners to prefer Post speech samples over Base samples were maintained at FUP.

Parent Perceptual Ratings

Parent perceptual ratings are displayed in the second graph of Figure 16. Visual inspection of the data identified several strong trends (i.e., 20% or more change) between study phases. Improved parent perceptions of loudness, monotone, breathiness, and understandability were identified from Base to Post. All of these improved perceptions were maintained for the Base to FUP comparison. No other strong trends were observed.

Parent responses to the 14 questions on the treatment rating form revealed a strong favorable impression of the treatment outcome for P2 and for the treatment approach (all ratings of 4 – quite a bit, or 5 - a lot).

Summary of Results for P2

P2 demonstrated improvements for all maximum performance tasks and for all measures of sustained phonation from Base to Post and Base to FUP. The values for HNR did not show strong trends; however, there was an average increase of 2.2 decibels from Base to FUP. P2 markedly increased vocal SPL, f_0 and f_0 range for sentence repetition and cartoon description across study phases. Changes in acoustic measures that moved P2 in the direction of performance of her typically developing peers occurred for all maximum performance tasks, vocal SPL and HNR values in sustained phonation, and vocal SPL in sentence repetition. For all other acoustic measures, there was either no change in performance or a change that moved in a direction away from the performance of her typically developing peers. Listener ratings of paired speech samples revealed a strong preference for treated speech samples (Post and FUP) over Base samples for nearly all perceptual variables. Finally, P2's parent ratings revealed improved perceptions of speech across study phases, and a favorable response to the treatment approach.

Data for P3

Maximum Performance Tasks

Maximum performance task data for P3 are displayed in the third graph of Figures 2 and 3. Grand means (and SDs) and mean differences for these measures are presented in Table 4. There are data for only one FUP session due to equipment failure. Visual inspection of maximum sustained vowel data revealed a strong increasing trend from Base to FUP. For maximum highest f_0 data, visual inspection revealed a slight decreasing trend from Base to Post (i.e., values were overlapping) and no trends from Base to FUP. Visual inspection of lowest f_0 data revealed a decreasing trend from Base to Post and Base to FUP (non-overlapping values). Visual inspection of maximum f_0 range data revealed a slight trend for decreasing f_0 range from Base to Post (values were overlapping), and no trends between Base and FUP.

<u>Sustained Phonation (Three Longest Vowels)</u>

Vocal SPL, f_0, and HNR data for P3 are displayed in the third graph of Figures 4, 5, and 6, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 4. Visual inspection of vocal SPL data revealed a strong increasing trend from Base to FUP. Mean f_0 and HNR data revealed no trends across study phases.

<u>Sentence Repetition</u>

Vocal SPL, f_0, and f_0 range data for P3 are displayed in the third graph Figures 7, 8 and 9, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 4. Visual inspection of vocal SPL, f_0, and f_0 range data revealed no trends across study phases for these measures.

<u>Cartoon Description</u>

Means and mean differences for vocal SPL, f_0, and f_0 range data for P3 are presented in Table 4. Data from cartoon description for FUP were not available due to microphone failure. Vocal SPL markedly decreased from Base to Post (7.4 dB SPL). Mean f_0 and f_0 range values also decreased from Base to Post.

<u>Listener Ratings</u>

Percentage of listener ratings for preference of speech samples spoken at Base, Post, and FUP are displayed in the third graph of Figures 10-15. Visual inspection of the data identified strong trends for Post speech samples over Base for the perceptual characteristics of loudness variability, pitch variability, overall voice quality, and articulatory precision. Overall voice quality was the perceptual variable for which listeners rated the strongest preference for Post speech samples over Base. Strong trends were maintained from Base to FUP for the perceptual characteristics of loudness variability, pitch variability, and overall voice quality. Increasing trends (i.e., less than 20% change) were observed for overall loudness and overall pitch across study phases. There were no strong trends for preferences between Post of FUP speech samples.

Parent Perceptual Ratings

Parent perceptual ratings are displayed in the third graph of Figure 16. Visual inspection of the data identified changes in parent perceptions for several variables rated. Strong trends (i.e., 20% or more change) were observed for improved perceptions of loudness, breathiness, strain, and frustration with talking from Base to Post. The improved parent perception of increased loudness was maintained from Base to FUP. No other strong trends were observed.

Parent responses to the 14 questions on the treatment rating form were all rated a 4 (quite a bit) or 5 (a lot) indicative of a favorable impression of treatment outcomes and the treatment approach.

Summary of Results for P3

Improvements in maximum performance tasks were observed for nearly all measures across study phases and for vocal SPL and HNR in sustained phonation. For speech tasks, there were no strong trends observed across study phases for sentence repetition and there was a decrease in values for all measures of cartoon description from Base to Post. Changes in acoustic measures that moved P3 in the direction of performance of his typically developing peers occurred for maximum performance tasks, vocal SPL and HNR values of sustained phonation, and vocal SPL, f_0, and f_0 range values of cartoon description. For all other acoustic measures, P3 either demonstrated no change in performance or a change that moved P3 away from the direction of performance of his typically developing peers. For listener ratings, there was a strong preference for treated speech samples (Post or FUP) as compared to Base speech samples for nearly all perceptual variables rated. Parent ratings on several perceptions of speech, voice and communication characteristics and impressions of the treatment technique were favorable at Post.

Data for P4

Maximum Performance Tasks

Maximum performance task data for P4 are displayed in the fourth graph of Figures 2 and 3. Grand means (and SDs) and mean differences for these measures are presented in Table 5. Visual inspection of maximum sustained vowel data revealed no strong trends across study phases. Due to this child's significantly reduced f_0 range, it was difficult to identify trends on the scale presented in this figure; therefore, data were expanded on a smaller scale for visual inspection of trends. Visual analysis of maximum f_0 data revealed an increasing trend from Base to Post and Base to FUP (i.e., accompanied by non-overlapping values). There is a missing data point for minimum f_0 for Base 3 due to the participant's unwillingness to perform the task. Visual inspection of minimum f_0 data revealed no trends across study phases. Visual inspection of maximum f_0 range data revealed a strong trend from Base to Post and Base to FUP.

Sustained Phonation (Three Longest Vowels)

Vocal SPL, f_0, and HNR data for P4 are displayed in the fourth graph of Figures 4, 5, and 6, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 5. Visual inspection of vocal SPL data revealed an increasing trend in baseline performance with no strong trends across study phases. Mean f_0 data revealed no strong trends across study phases. Visual analysis of HNR data revealed a strong increasing trend from Base to FUP.

Sentence Repetition

Vocal SPL, f_0, and f_0 range data for P4 are displayed in the fourth graph of Figures 7, 8, and 9, respectively. Grand means (and SDs) and mean differences are presented in Table 5. P4 was unable to repeat entire sentences; therefore, he repeated the last word of each sentence presented ("puppy", "pot", "key"). Data for this task are from these single word repetitions. There are no data for Base 2 due to P4's unwillingness to perform the task, and no data for FUP 2 due to microphone failure. Visual inspection of vocal SPL data revealed an increasing trend from Base to FUP (non-overlapping values). Mean f_0

data revealed no trends across study phases. Visual inspection of f_0 range data revealed an increasing trend from Base to FUP (i.e., accompanied by non-overlapping values).

Listener Ratings

Percentage of listener ratings for preference of speech samples spoken at Base, Post, and FUP sessions are displayed in the fourth graph of Figures 10-15. Pair comparisons for P4 were of three repetitions of single words ("puppy", "pot", "key"). Visual inspection of the data identified strong trends for preference of Post speech samples over Base for all perceptual variables. Overall voice quality was the perceptual variable for which listeners had the greatest preference for Post over Base speech samples. Strong trends in preference for FUP speech samples over Base were observed for all variables except overall pitch and pitch variability where there were trends in data, but not strong ones. There were no strong trends for preference of Post over FUP speech samples for any perceptual variables rated.

Parent Perceptual Ratings

Parent perceptual ratings are displayed in the fourth graph of Figure 16. Parent ratings were only available for Base and Post. Visual inspection of the data identified changes in four parent perceptual ratings from Base to Post. Strong increasing trends were observed for perceptions of loudness and frustration with talking, and strong decreasing trends were observed for hoarseness and initiating talking/conversations. No other strong trends were observed.

Parent responses to the 14 questions on the treatment rating form were all rated a 4 (quite a bit) or 5 (a lot) indicating a favorable impression of P4's treatment outcome and of the treatment approach.

Summary of Results for P4

For maximum performance tasks, P4 demonstrated improved performance for all values with strong trends for maximum highest f_0 and f_0 range. HNR values of sustained phonation improved, on average, 8.9 decibels from Base to FUP. A strong trend for increased f_0 range in speech was observed from Base to FUP. Changes in acoustic measures across study phases that moved P4 in the direction of performance of his typically developing peers occurred for maximum performance tasks, vocal SPL and HNR values of sustained phonation, and vocal SPL and f_0 range of sentence repetition. For all other acoustic measures, P4 either demonstrated no change in performance or a change that moved P4 in a direction away from the performance of his typically developing peers. For listener ratings, there was a strong preference for treated speech samples (Post and FUP) over Base samples. Changes in parent perceptual ratings included improved and worsened perceptions, but overall impression of the treatment approach was favorable.

Data for P5

P5 was an untreated comparison participant in this study; therefore, the study phases do not include a Post or FUP phase. To distinguish among baseline sessions for P5, Base A will represent the initial 4 baseline sessions within 3 weeks time. Base B will represent the two baseline sessions one month after the last session in Base A. Base C will represent the two baseline sessions 6 weeks after the last session in Base B.

Maximum Performance Tasks

Maximum performance task data are displayed in the fifth graph of Figures 2 and 3. Grand means (and SDs) and mean differences for these measures are presented in Table 6. Visual inspection of maximum sustained vowel data revealed no strong trends across study phases. For maximum highest f_0, a strong decreasing trends from Base A to Base B, and to a lesser extent from Base A to Base C were observed (non-overlapping values). Minimum lowest f_0 data revealed no trends across study phases. Maximum f_0 range data revealed a strong decreasing trend from Base A to Base B.

Sustained Phonation (Three Longest Vowels)

Vocal SPL, f_0, and HNR data for P5 are presented in the fifth graph of Figures 4, 5, and 6, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 6. Visual inspection of vocal SPL, f_0, and HNR data revealed no trends across study phases for any of these measures.

Sentence Repetition

Vocal SPL, f_0, and f_0 range data of sentence repetition for P5 are displayed in the fifth graph of Figures 7, 8, and 9, respectively. Grand means (and SDs) and mean differences for these measures are presented in Table 6. Visual inspection of vocal SPL, f_0, and f_0 range data for sentence repetition revealed no strong trends across study phases for these measures.

Cartoon Description

Means (and SDs) and mean differences for vocal SPL, f_0, and f_0 range data are presented in Table 6. For vocal SPL, there was a slight decrease of 1.7 decibels from Base A to Base B, and no difference between Base A and Base C. There was minimal change for f_0 across study phases. There was a small increase from Base A to Base B for f_0 range (14 Hz), followed by a decrease in f_0 range (57 Hz) from Base A to Base C.

Listener Ratings

Percentage of listener ratings for preference of speech samples spoken at Base A, Base B, and Base C are presented in the fifth graph of Figures 10-15. Visual inspection of the data revealed no trends for preference of one Base condition (A, B, or C) over another. The small percentages for preferences across all perceptual variables indicate that most often listeners made a choice of "no preference" as opposed to choosing one sample over another. These data suggest that P5's speech and voice characteristics did not change over the course of the study in a way that influenced listener preference on the six perceptual variables rated.

Parent Perceptual Ratings

Parent perceptual ratings are displayed in the fifth graph of Figure 16. Parent ratings were only available for Base A and Base B. Visual inspection of the data identified a strong increasing trend for the perception of hoarseness and a strong decreasing trend for the perception of speaking so others can understand. No other strong trends were noted.

Summary of Results for P5

P5 did not demonstrate strong trends for increased performance on maximum performance tasks or sustained phonation measures. In contrast, there was an observable decreasing trend for maximum f_0 and f_0 range. There were minimal changes in acoustic measures for the sentence repetition and cartoon description tasks across study phases. Changes in acoustic measures across study phases that moved P5 in the direction of performance of her typically developing peers occurred for f_0 range of cartoon description from Base A to Base C. For all other acoustic measures, P5 either demonstrated no change or a change that moved her away from the direction of performance of her typically developing peers. Listener preference ratings did not reveal any strong trends for preference of speech samples across Base A, B, and C. Finally, parent ratings revealed minimal changes in perception of speech and voice characteristics.

Patterns of Results Across Participants with CP

A few general patterns of performance across participants with CP who received treatment were observed. The majority of strong trends revealed in acoustic measures were for maximum performance tasks (which were training targets in therapy). All participants made substantial gains in one or more of these measures and maintained or increased gains in acoustic measures over time. Consistent strong trends were not as frequently observed for sustained phonation or speech tasks (with the exception of P2 who demonstrated strong trends on almost all measures). For all participants with CP, HNR values were below the mean and outside the range of performance of their typically developing peers across all study phases. This was the only variable where no participants with CP even reached the range of performance of their typically developing peers at some phase in the study. Participants 1-4 who received treatment did, however, show a consistent trend for increased HNR values across study phases. P5 demonstrated no change over time for this measure. All participants with CP had a mean fundamental frequency on speech tasks that was above the mean, and in many cases, outside the range of performance of typically developing peers. The higher fundamental frequency for speech did not change across study phases for the treated participants, and in some cases the fundamental frequency was even higher at Post and FUP as compared to Base.

There was an overwhelming preference on the part of listeners for speech samples from Post and FUP as compared to Base in the participants with CP who received treatment. The exception was P1, where Base speech samples were more preferred than FUP samples. All parents of treated participants with CP reported improved perception of loudness from Base to Post ratings. Parent ratings of other perceptual variables varied across participants. Finally, all parents of treated CP participants rated "agree" or "strongly agree" to the 14 questions related to perceptions of treatment, suggesting a favorable response to treatment.

DISCUSSION

The purpose of this study was to examine the effects of an intensive voice treatment in children with spastic CP. The two research questions posed by this study were, Will intensive treatment perturb the speech motor system of children with spastic CP? and, If speech is perturbed, is there a therapeutic effect? All participants who received treatment (P1, P2, P3, P4) demonstrated positive changes in selected acoustic measures post-treatment. These changes were accompanied by listener preference for speech samples from the post-treatment condition over baseline samples. There was minimal change in the control participant (P5) over time. These findings suggest that the speech system was perturbed and that a therapeutic effect was detected in participants with spastic CP following intensive voice therapy. The implications of these findings and directions for future research are discussed below.

Perturbation of the Speech Motor System

The greatest and most consistent changes in acoustic measures occurred in maximum performance voice tasks of participants with CP who received treatment (P1-P4). This finding is not surprising as these were training tasks in treatment sessions. When compared to the data of typically developing peers in this study, participants with CP were outside the range of performance for all maximum performance tasks across study phases (i.e., even after treatment). These findings are consistent with those of Wit et al. (1993) who reported that children with spastic CP were significantly reduced in their ability to perform maximum tasks as compared to their typically developing peers. Hardy (1967) has suggested that children with CP have a combination of speech system limitations that would predict decreased maximum performance capabilities and speech breathing difficulties. These limitations include a reduced vital capacity combined with weak and incoordinated breathing musculature, and poor laryngeal and upper airway valving (Hardy 1967; Solomon & Charron, 1998). Despite these presumed speech system limitations, the four participants who received treatment were able to increase their maximum performance values after treatment, moving them closer to the range of performance of their typically developing peers.

The value of training maximum performance tasks (voice or speech) in treatment of children with CP has been questioned. Hardy (1983) argued against using these tasks for two reasons: (a) the risk of motor overflow with increased effort, and (b) because speech seldom involves activities that require maximum effort. Hardy cautioned that stimulating high effort in children with CP may result in unwanted associated movements (i.e., effort directed to speech musculature would overflow to limb and body musculature). The present participants exhibited some associated movements (facial grimacing, extension of the body, arms squeezing against the chest) with stimulation of increased effort at the beginning of treatment; however, these movements decreased or were eliminated over time, as judged by the investigator and parents. Repeated motor practice trials with a focus on sensory awareness and intensive training may have facilitated specificity of muscle activity to the degree that effort directed to the speech system did not spread across other systems (limb movements). This hypothesis is consistent with findings from studies in adult monkeys where behavioral conditioning tasks that incorporated repetitive sensory inputs and movements resulted in progressively more refined and differentiated cortical representations of muscle, joint afferent fibers, and motor movements (Byl et al., 1996). These findings are also consistent with reports in limb literature that children with CP are able to achieve isolated muscle strength through resistance training programs (Damiano, Vaughn et al., 1995) and can benefit from maximum effort exercises with no increase in spasticity in other parts of the body (Fowler et al., 2001).

Hardy's (1983) second argument against maximum performance tasks is that speech seldom involves activities that require maximum effort. Even so, this does not diminish the potential benefit children with CP may receive from maximal effort training. It has been well-documented that children with CP fatigue more easily than their typically developing peers (Brunstrom, 2001); yet, the source of fatigue is unclear and may be the result of a sedentary lifestyle. The impact of a sedentary lifestyle on speech had not been specifically studied, but many children with CP produce even simple conversational speech with great effort, while experiencing physical and vocal fatigue. Typically developing children are "vocal athletes" by contrast. They have vast experiences of running, skipping, hopping while phonating or talking. They manipulate postural positions from standing up straight to standing on their heads and learn to make adjustments to control speech. These experiences may facilitate their ease with speaking. Lack of such experiences in children with CP (coupled with motor impairments in speech musculature) may exacerbate their difficulties with speech production.

In treatment, maximum performance tasks for participants with CP were used to simulate high respiratory/phonatory energy expenditure that typically developing children may experience from moving and speaking. Goals in treatment were to provide opportunities for participants with CP to overcome effort and fatigue with speaking by pushing them to a target vocal SPL (in voice and speech tasks) and requiring them to maintain that target, longer and longer (i.e., increased duration vowels, from words, to phrases, to conversation), and over and over again (i.e., repeated practice trials). As a result, fatigue and physical effort with speaking appeared to diminish over time, and participants with CP were perceived by the investigator, parents, and listeners to produce conversational speech with less effort. In addition, maximum performance tasks were key in facilitating respiratory/laryngeal coordination (e.g., learning to time vocal fold adduction with expiration), training of good vocal quality (e.g., decreased strain and breathiness), building consistency and stability of the voice (e.g., maintain target vocal SPL for longer periods of time), and endurance (ability to go longer and more repetitions without vocal or physical fatigue).

It is not surprising that changes in acoustic measures for speech tasks were not as large as in maximum performance voice tasks as there are acceptable limits to vocal SPL, f_0 and f_0 range targets for speech (i.e., not too loud, too high/low, or too variable). At baseline, some participants with CP were able to achieve vocal SPL equal to typically developing peers, but the quality of the voice was often strained or pressed. Other patterns of behavior of the participants with CP included speaking with adequate loudness or excessive loudness at the start of a sentence and then trailing off into whispered speech. Thus, large absolute changes in acoustic measures were not always required; rather, learning to maintain and modulate vocal SPL and f_0 with good vocal quality in continuous speech production was the goal.

Evidence for a Therapeutic Effect

To determine if perturbation of the speech motor system resulted in a therapeutic effect, listeners rated their preference for speech samples from participants with CP across different study phases. Overall, listener ratings revealed strong preferences for post-treatment speech samples over baseline samples on nearly all perceptual variables rated for all four treated participants (P1-P4). Listeners were encouraged to provide comments related to their choice of preference, which provided additional insight into perceived changes in speech and voice functioning after treatment.

Listeners commented that they preferred post-treatment speech samples over baseline samples because they were louder, there was consistent voicing throughout the sentences (i.e., less whispered speech), and the voice sounded "stronger." Comments related to post-treatment preferences also indicated that pitch was more natural, comfortable, and less monotonous, and voice quality was perceived as less strained, less glottal fry, and not as breathy or hoarse. Listeners preferred post-treatment articulation because it was perceived as more "crisp," words didn't "run together as much," and articulation was more "precise and faster." General comments related to overall effort were also reported, such as speech was "less of a struggle," speech sounded "less effortful," and a perception that it was "easier" for participants to produce speech. One notable exception was for P2 who was yelling in a follow-up speech sample (listeners preferred baseline and post-treatment samples because they were softer and not "screaming"). This yelling behavior was not trained in treatment.

Parent perceptual ratings matched listener ratings for participants, except for P4 where parent ratings revealed an increased perception of "hoarseness" post-treatment. This may be explained by the fact that after therapy P4 was talking more and in longer phrases, which may have made parents more aware of certain voice qualities. This rating did not fit with listener ratings of an improved voice quality for P4 post-treatment.

The perceptual comments of a louder, stronger voice, improved control over pitch and loudness variability, decreased strain and breathiness, and more precise articulation all support the concept that training increased vocal effort and loudness can have a global positive impact on speech (Dromey & Ramig, 1998a; 1998b; Dromey et al., 1995; Fox et al., 2002; Ramig, 1992; Ramig & Dromey, 1996). McClean and Tasko (2002) recently reported evidence for neural coupling of orofacial muscles to neural systems of laryngeal and respiratory control. These authors suggest that a potential source of this observed neural coupling may be from efferent drive from a common brain region to motoneurons innervating orofacial, laryngeal, and respiratory muscles. Neural coupling may explain, in part, the potential spread of effects from stimulation of increased vocal effort and loudness (respiratory and laryngeal systems) to orofacial muscles. Schulman (1989) suggested as well that targeting loud speech may result in a reorganization of motor activity that is not restricted to respiratory and phonatory mechanisms, but includes increased articulatory movement. Accordingly, increased neural drive to orofacial muscles has been associated with increased vocal effort (McHenry, 1997; Ramig, et al., 2000; Wohlert & Hammen, 2001) and may have contributed to the perception of improved articulatory precision in this study (when articulation was not directly treated in therapy). McHenry (1997) and Young et al. (2001) reported that increased vocal effort may also directly facilitate velopharyngeal closure (depending upon severity of velopharyngeal impairment). For P1, there were comments of decreased nasality and fewer audible nasal emissions associated with post-treatment speech samples. Dromey and Ramig (1998) suggested that maximum sustained vowel tasks performed at increased vocal effort and loudness levels likely makes the muscles of phonation become stronger and steadier in their function, and increased loudness has been associated with more stable motor output (Klienow et al., 2001). These observations may explain listener comments that voice and speech sounded "stronger" and "more consistent." In addition, speaking with increased vocal effort may portray greater confidence or other personality factors that could have influenced listener preference in this study (Lienard & Di Benedetto, 1999). Thus, the potential that a single treatment target (i.e., increased vocal effort and loudness) may impact multiple speech subsystems is an important consideration for delivering efficient treatment to children with spastic CP.

Did Therapeutic Effects Last?

To determine if the effects of treatment lasted over time, follow-up voice recording sessions were conducted 6 weeks after the conclusion of treatment for participants with CP. Three participants who received treatment (P2, P3, P4) maintained or continued to improve performance across nearly all acoustic measures and tasks at follow-up. P2 appeared to embrace the concept of using her "strong voice." Parent reports indicated that P2 frequently reminded herself to use her strong voice and she used it regularly in daily communication. P3 needed some cueing to use his strong voice in daily communication. P3's mother reported that if she could not understand him, cueing him to repeat with his "strong voice" was a helpful strategy. In some cases she reported that he would cue himself to use his "strong voice" or tell other people to use a strong voice if he could not understand them. For P4, parent comments suggested that he continued to practice and improve on his ability to use treatment techniques after the conclusion of therapy. P4 had four older siblings who routinely participated in treatment sessions during the one month of therapy. These siblings continued to work with P4 at the conclusion of treatment, and constantly cued him to use his "big voice." It is likely that this family support contributed to P4's maintenance of treatment effects over time.

For P1, continued improvements from post-treatment to follow-up were observed for maximum performance tasks; however, changes in speech were not maintained. Listener preference ratings rarely chose a follow-up speech sample over post-treatment or baseline samples. Also, parent ratings suggested that treatment effects had diminished over time. The parent reported this was due, in part, to P1's unwillingness to practice on his own, or with family members. The parent felt there needed to be an external person (e.g., speech therapist) to work with her child. Throughout therapy, P1 displayed a persistent resistance to using his "strong voice" outside of the home environment. He expressed concern about talking louder at school, saying, "the other kids will think I am crazy," although he admitted that his "strong voice" was no different than the speech of his typically developing peers.

The ability of participants in this study to maintain treatment effects appeared to depend upon multiple factors, such as acceptance of the treatment techniques, social and emotional factors, and family support. Bower and McLellan (1992) anecdotally reported that motor goals achieved in physical therapy by children with CP were maintained and even continued to improve over time if the motor action learned in treatment was incorporated into daily living. Future studies are required to determine duration of treatment effects associated with intensive voice therapy (e.g., more than 6 weeks) and to delineate variables that contribute to long term treatment success.

General Comments on the Treatment Model

The treatment approach used in this study differs from traditional speech treatment approaches for children with CP. This approach conforms to current theories of motor development and principles of motor learning as summarized below. Thelen (1996) has suggested that movements in atypical populations (e.g., children with CP) are not obligatory patterns resulting from a primary deficit of the central nervous system (CNS); rather, they are patterns selected by the CNS to accomplish goals, such as moving to a target or speaking. If the motor system has used selected patterns for many years, then motor habits can be difficult to change. Latash and Anson (1996) suggest that a damaged nervous system will search for optimal strategies and eventually may reach a local minimum of a function (set pattern of achieving a goal, such as speaking) and remain there because deviating from this minimum degrades the optimized behavior (i.e., deviating from current pattern of speech will result in degraded speech). As a result, the system may "... never be able to discover that, beyond the nearest edge, there is another, much deeper minimum corresponding to a much better value of the optimized function.

A therapist may know this and may persuade the patient (and the CNS) to continue the search even when it leads through apparent discomfort" (Latash & Anson, 1996, p. 98).

The treatment used in this study attempted to push participants beyond their current level of functioning using an intensive voice treatment, that incorporated high effort, repeated practice trials, and sensory awareness/sensory augmentation training (Fox et al., 2002; Ramig, Countryman, et al., 1995; Ramig, Pawlas, et al., 1995; Ramig et al., 1996). This treatment focused specifically on voice (i.e., increasing vocal effort and loudness, with good vocal quality) to improve speech. The target of loud/strong/big voice appeared cognitively easy for the present participants to understand, and they were all able to perform treatment tasks. Despite the fact that increased tone and spasticity are motor signs in children with spastic CP, the high effort, intensive treatment did not appear to exacerbate these signs in any of the participants. These findings are consistent with reports from Fowler et al. (2001) and Damiano, Vaughan et al. (1995) who reported no increased spasticity following high effort exercises in limb training. For participants who demonstrated strain-strangle or pressed voice quality during baseline sessions, the treatment did not make them worse. In fact, acoustic measures (i.e., HNR) and listener ratings indicated that voice quality improved. It may be that poor voice quality prior to treatment was related to generalized weakness of respiratory and laryngeal musculature, coupled with incoordination of respiratory/laryngeal subsystems. With treatment, improved strength, endurance, and coordination eliminated the need for these hyperfunctional behaviors and replaced them with good voicing quality. This is similar to findings in individuals with Parkinson disease after LSVT (Countryman et al., 1998; Smith et al., 1995).

Intensive treatment was well-tolerated by all participants, with 100% compliance from participants and families to the treatment regime. In addition, all families (except the family of P4) enjoyed the intensive model. These findings are similar to those of Schindl et al. (2000) who reported eight of ten children and families enjoyed an intensive gait training program. Although several of the participants in this study demonstrated some physical fatigue at the start of treatment (e.g., parents would report that after treatment the child took a nap, or was "wiped out" for the evening), over time this fatigue diminished and participants were able to easily work through treatment session without fatigue later in the day. P1's parent reported that after treatment he was "energized" and performed better on other tasks, such as physical therapy sessions on that same day. Dosage of intensive therapy may have been an important factor in this study. The 1 month of intensive treatment in this study was acceptable to participants and families and resulted in some positive effects; however, future studies are needed to determine the optimal dosage of therapy for maximum treatment outcomes.

Repeated practice trials in therapy accompanied by cues to increase vocal effort were also well-tolerated by participants with CP. In this treatment, repetitions of tasks were conducted while encouraging participants to actively attend to the sensory feedback that accompanied motor output. This active attention to sensory feedback may be essential for facilitating neuroplastic changes of cortical sensorimotor maps during behavioral treatment (Byl et al., 1996; Lenz & Byl, 1999). For example, treatments for patients with dystonias that incorporate active sensory awareness training with movement have shown promise for cortical reorganization of sensorimotor maps resulting in improved motor control, motor accuracy, and sensory discrimination (Byl & McKenzie, 2000). In contrast, repetitive passive inputs without a sensory focus, which were common activities in the early NDT approach, have been shown to generate minimal enduring neuroplastic changes (Byl et al., 1996; Byl & McKenzie, 2000; Lenz & Byl, 1999). The degree to which the damaged nervous system of a child with CP can reorganize cortical maps to improve functional behavior is not clear. Nevertheless, including treatment strategies that have been documented to maximize the potential for neuroplastic change may be important.

Sensory augmentation (e.g., high effort levels) and sensory awareness (e.g., "Do you feel that effort?") of respiratory and phonatory requirements for speaking in participants with CP improved task performance and may have contributed to motor learning (Schmidt & Lee, 1999). For example, some participants with CP stopped voicing during sustained vowel tasks, apparently without a physical understanding of what it meant to keep going "until all their air was gone." Sensory augmentation exercises such as bouncing the participant on a therapy ball while phonating, and touching their voice box while encouraging them to "go, go, go" seemed to raise their awareness of respiratory/phonatory effort and facilitated the participant's maximum performance. Later in the course of treatment, participants were able to perform maximum performance tasks without the need for sensory augmentation or continued cueing from the investigator. This observation suggests that participants may have developed an internal representation or understanding of maximum effort enabling them to achieve this target on their own. Sensory awareness activities also targeted auditory and cognitive awareness (i.e., as opposed to physical awareness) in participants with CP by using tape recorders to allow participants to hear their own voice, making phone calls, and input from parents and sibling regarding the "goodness" of their strong voice.

Parents reported a favorable response to the treatment, which may have facilitated participant compliance in this study. For example, parents perceived the assignments as easy because the participant could do them while the parent was doing something else (e.g., driving in the car). All parents reported that they felt their child had fun participating in the treatment and that the treatment techniques were something their child was able to learn and incorporate into his or her daily living. All parents reported that they would recommend this type of treatment to other families who have children with CP. One parent suggested that she would have liked to seen some peer group interaction incorporated into the treatment approach. The investigator agrees that this may be essential to facilitate maximum impact of the treatment techniques in daily living.

Study Critiques and Future Directions

This study represented a Phase 1 research design to examine potential therapeutic effects associated with an intensive treatment approach for children with spastic CP (Robey & Schulz, 1998). A single subject design was used and therefore results of this study can not be generalized to the population of children with spastic CP. The results of this study revealed that participants who received treatment were able to make improvements in several acoustic measures, and that a positive therapeutic effect lasted over time. These findings provide support for further examination of the effects of high effort, intensive treatment with a sensory focus on speech of children with spastic CP.

Several design flaws in this study were noted including the number of recording sessions, data collection tasks, and carry over exercises in the treatment model. Given the nature of variable performance for children with spastic CP, more than two recording sessions at post-treatment and follow-up would have been valuable. A third data point may have offered a definitive picture of the child's post-treatment and follow-up variability as compared to baseline.

Some of the participants with CP were not as adept at spontaneous use of treatment techniques (e.g., strong voice), instead they required cueing. It would have been useful to elicit cued loud speech in baseline recording sessions (one task that is conducted at habitual and loud levels) in order to compare potential changes in cued speech at post-treatment and follow-up.

The task for spontaneous speech samples (cartoon description) was adequate, but there needed to be greater control over eliciting retelling of the cartoon in recording sessions. In some sessions the participant told the cartoon to a parent, in other sessions a sibling, and the listener may have influenced the child's behavior (e.g., P3 was fighting with his brother during the task). Having an external listener (not parent or sibling) who always elicited the retelling of the cartoon for every participant would have eliminated this inconsistency. In addition, standard probing questions and continuing the task until a standard duration of conversation was collected from every participant would also improve consistency of performance across participants on this task.

Regarding the treatment model used in this study, this population may require additional external resources to maximize carry over of treatment techniques outside of therapy sessions (i.e., not rely on the child to spontaneously use "strong voice" with others). Training teachers, school aides, and the child's other therapists to cue the child periodically to use his or her strong/big/loud voice would provide additional stimulation in a variety of social environments. In addition, training a few peers in the classroom of children with CP who could cue and reinforce their strong/big/loud voice may facilitate comfort and ease with using treatment techniques.

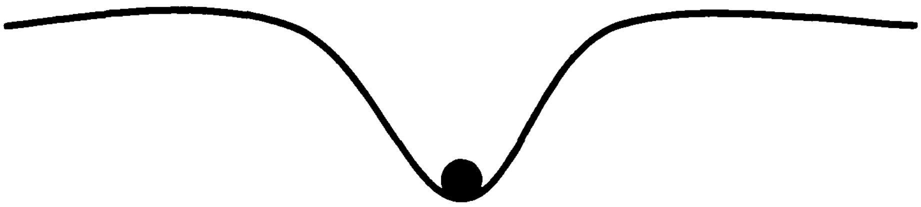
Summary and Conclusions

The purpose of this study was to explore the effects of an intensive speech treatment on perturbing the speech motor system of children with spastic CP, and to determine a therapeutic effect, if any. Acoustic measures related to voice functioning, auditory-perceptual analysis of speech samples, and perceptual ratings by parents of participants in this study were used. All four participants who received treatment in this study demonstrated a perturbation (marked change in performance) on one or more acoustic measures of voice and speech output. There were strong listener preferences for the treated speech samples (post-treatment or follow-up sessions) over baseline samples for most perceptual characteristics rated in this study. All parents reported improved perceptions of two or more voice, speech, or communication characteristics rated following treatment, and all had an overall favorable impression of their child's treatment outcome and of the treatment approach used. The participant with CP who did not received treatment demonstrated minimal changes in acoustic measures over time (and even a marked decrement in performance on select measures) and listeners did not show strong preferences for speech samples from one study phase over another. These findings suggest that in these four participants with CP, intensive treatment both perturbed the speech motor system and demonstrated a therapeutic effect. In addition, some measures of treatment effects were maintained for all participants 6 weeks after the conclusion of treatment.

Future studies may investigate potential physiological changes that may accompany treatment. In addition, examination of alternative treatment approaches administered in a parallel mode and intensity, but with a different focus (e.g., oromotor exercises, articulation) will help delineate key elements of treatment success. For example, was it the focus on vocal effort and loudness, intensity of treatment, or sensory activities that contributed most to treatment outcomes. Finally, studies that engage a larger number of children with spastic CP are required to determine generalizability of treatment outcomes for this population.

Figure 1. This figure represents attractors with wells of varying depths as described in dynamic systems theory. The depth of the well represents the strength of an attractor state. The filled circle (ball) in the well represents a behavior that may move in and out of an attractor state. The depth of an attractor well may determine the degree of difficulty to move a behavior (i.e., move the ball) from one state of functioning to another. (A) A deep attractor well representing a behavior that is stereotypical, almost obligatory. A great deal of energy may be required to move or change a behavior (i.e., move the ball) from such a deep attractor state. (B) A shallow attractor well representing a preferred, but flexible behavior pattern. Less energy may be required to move or change a behavior (i.e., move the ball) from this state into another attractor state.

A



B

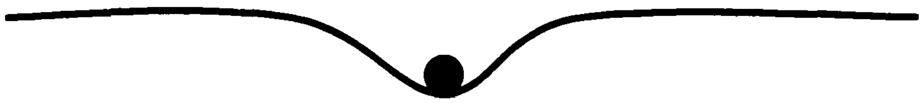
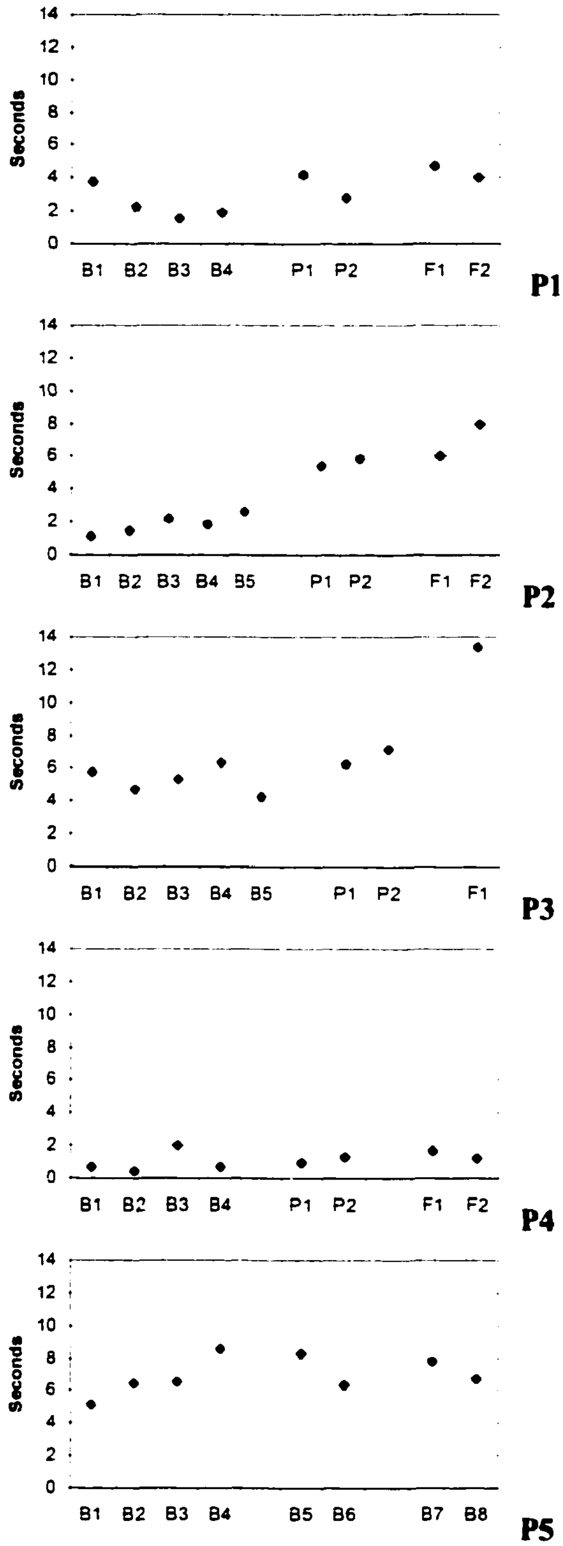


Figure 2. Maximum duration (longest) sustained vowel for Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the value of the single longest sustained vowel for a given recording session.

Figure 2. Maximum duration (longest) sustained vowels.



<u>Figure 3. Maximum highest and lowest frequency and frequency range across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). The top circle represents the highest frequency, the bottom circle represents the lowest frequency, and the connecting line represents the total maximum frequency range for each recording session.</u>

Figure 3. Maximum frequency range of sustained vowels.

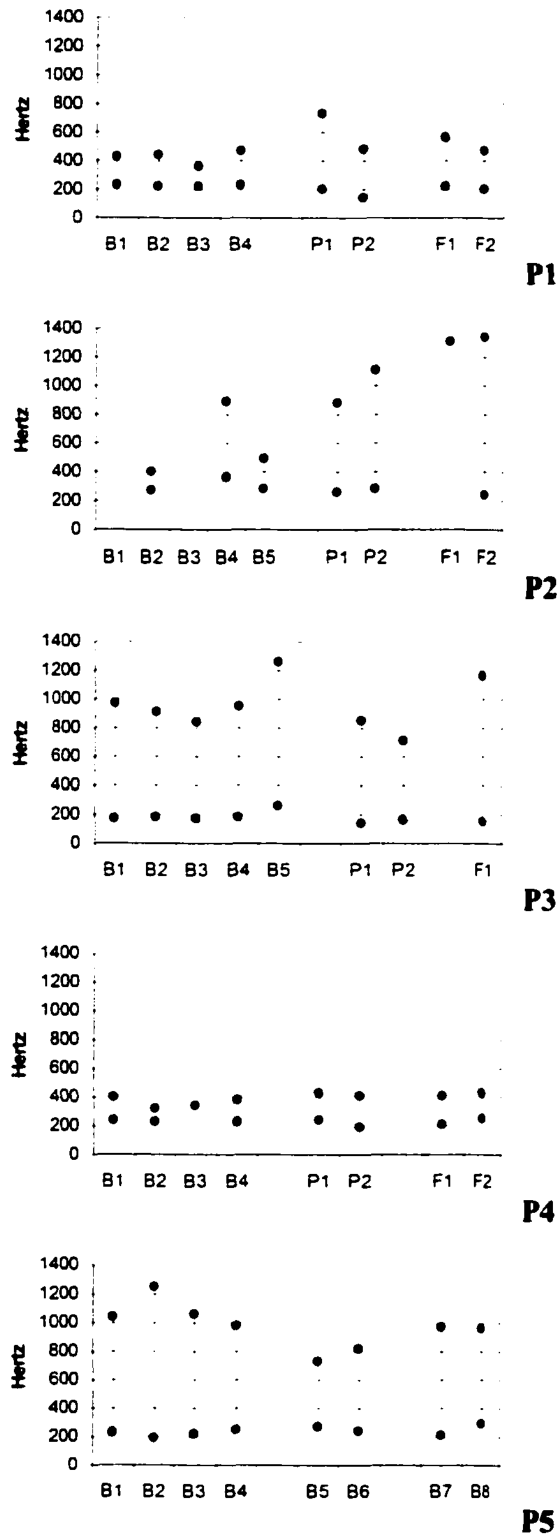


Figure 4. Mean vocal SPL (dB SPL at 4 inches) of the three longest sustained vowels across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the mean vocal SPL of the three longest sustained vowels per recording session. Error bars represent the standard deviation of these mean values.

Figure 4. Vocal SPL of three longest sustained vowels.

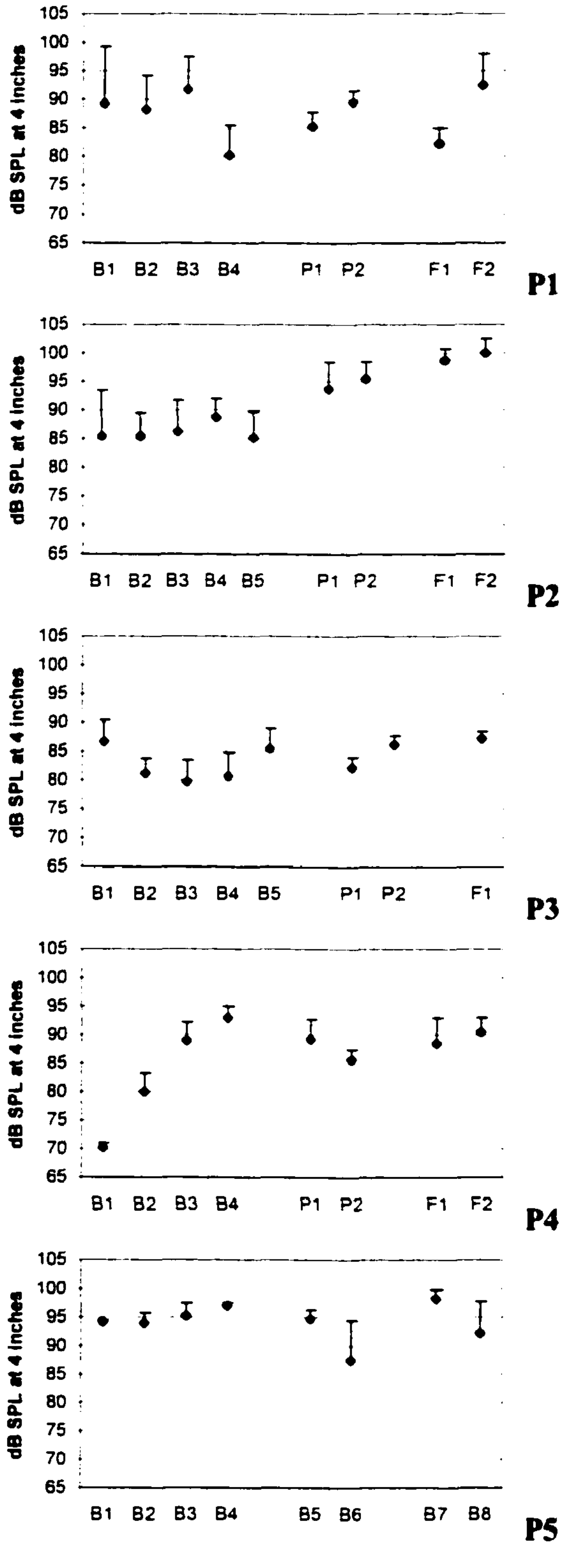
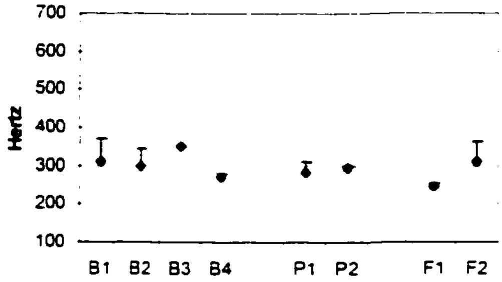
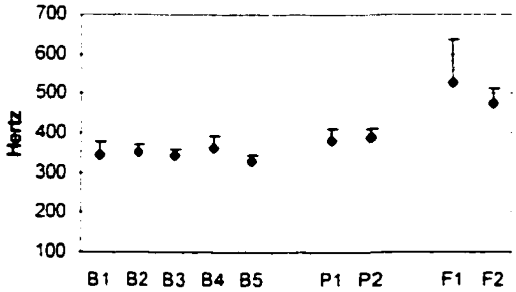


Figure 5. Mean fundamental frequency of the three longest sustained vowels across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the mean fundamental frequency of the three longest sustained vowels per recording session. Error bars represent the standard deviation of these mean values.

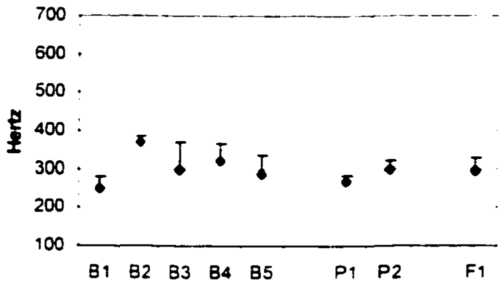
Figure 5. Mean fundamental frequency of three longest sustained vowels.



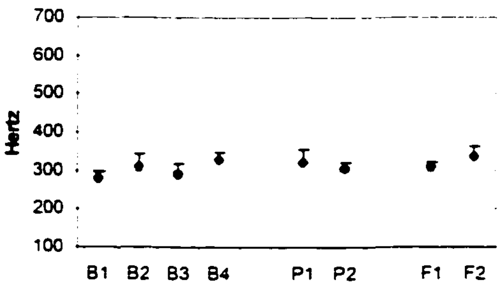
P1



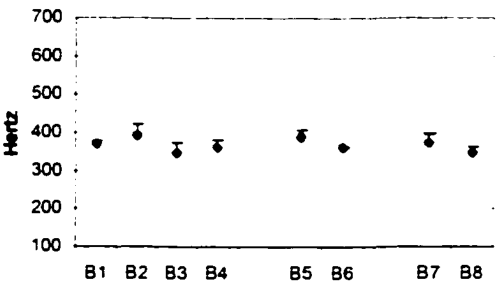
P2



P3



P4



P5

Figure 6. Mean HNR of the three longest sustained vowels across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the mean HNR of the three longest sustained vowels per recording session. Error bars represent the standard deviation of these mean values.

Figure 6. Mean HNR of three longest sustained vowels.

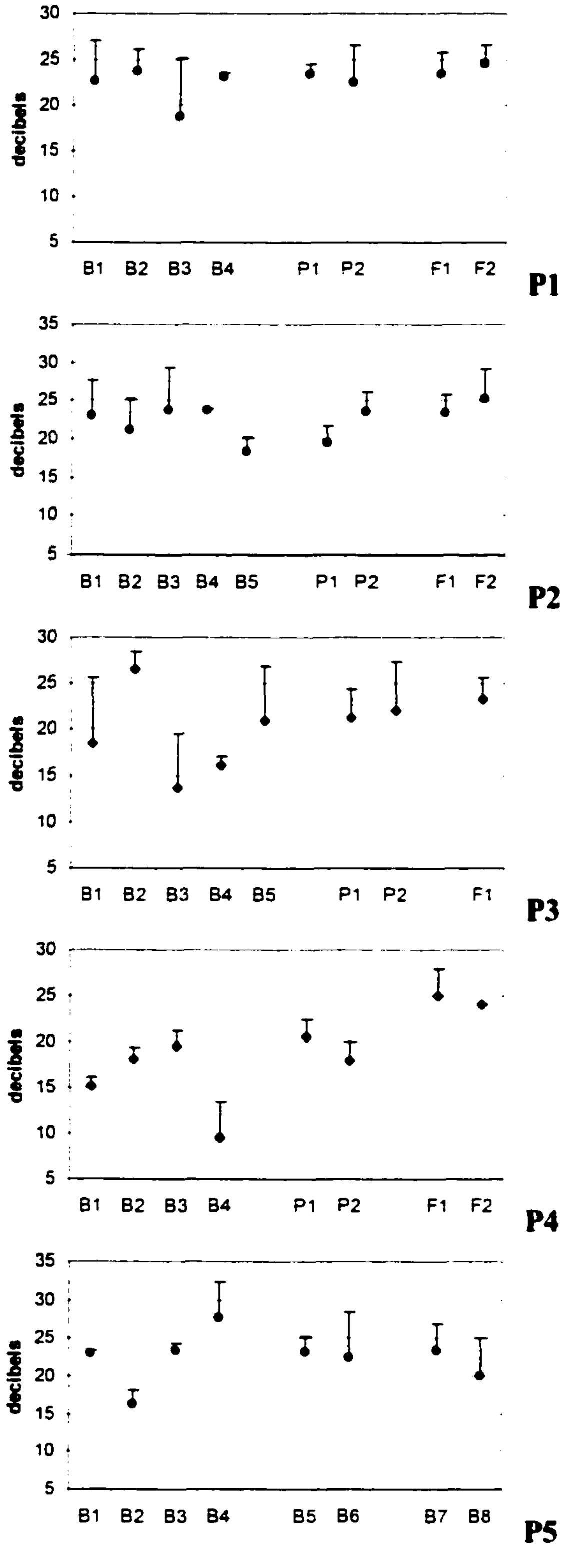


Figure 7. Mean vocal SPL of nine sentence repetitions across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the mean value for vocal SPL of nine sentence repetitions per voice recording session. Error bars represent the standard deviation of these mean values.

Figure 7. Mean vocal SPL of sentence repetition task.

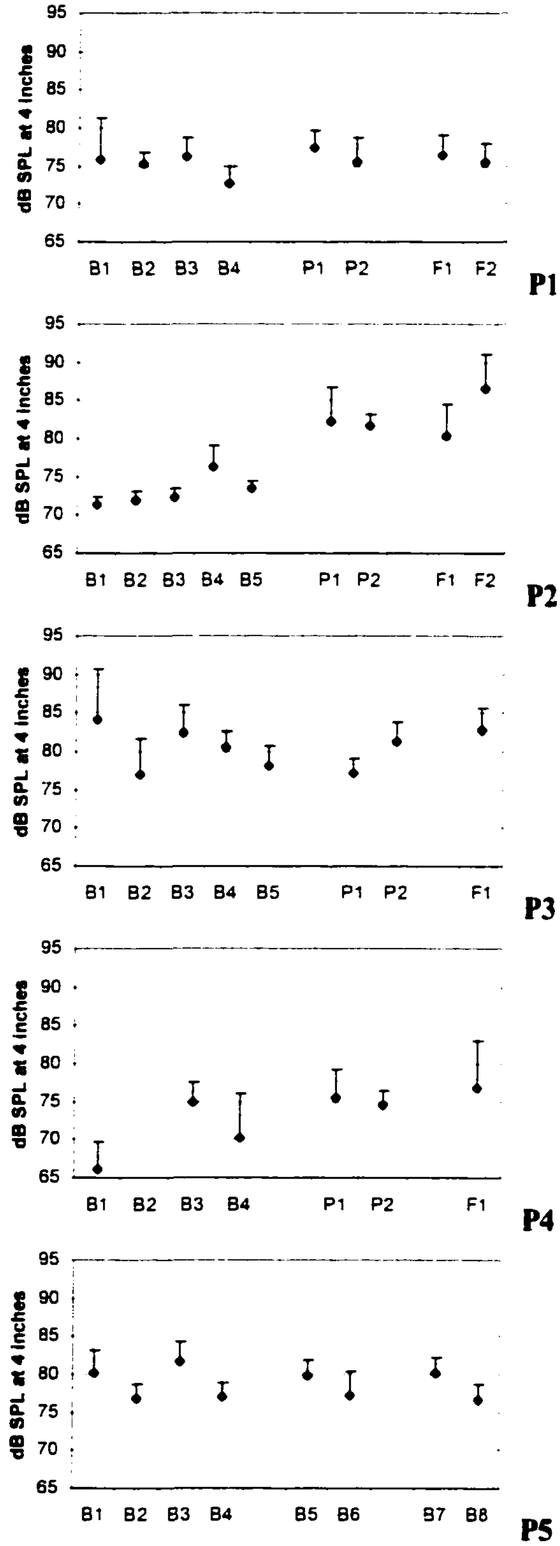
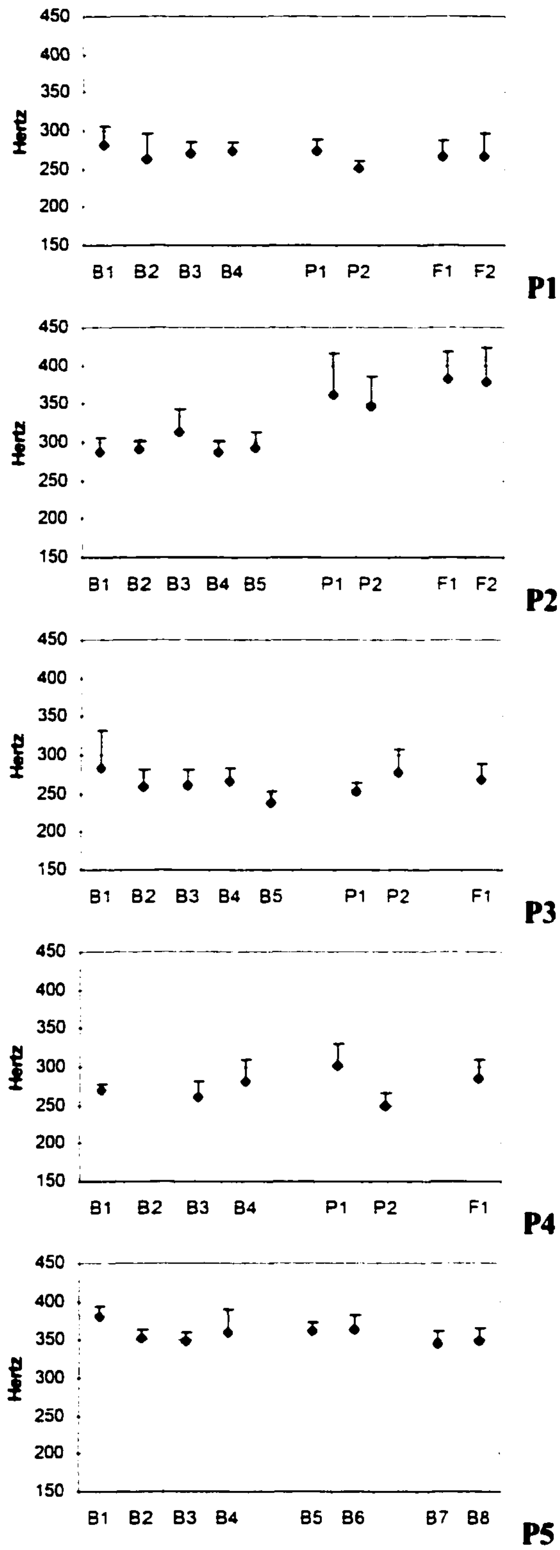


Figure 8. Mean fundamental frequency of nine sentence repetitions across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the mean fundamental frequency of nine sentence repetitions per recording session. Error bars represent the standard deviation of these values.

Figure 8. Mean fundamental frequency of sentence repetition task.



<u>Figure 9. Mean frequency range of nine sentence repetitions across Base (B), Post (P), and FUP (F) recording sessions for each participant (P1-P5). Each filled circle represents the mean frequency range of nine sentence repetitions per recording session. Error bars represent the standard deviation of these values.</u>

Figure 9. Mean fundamental frequency range of sentence repetition task.

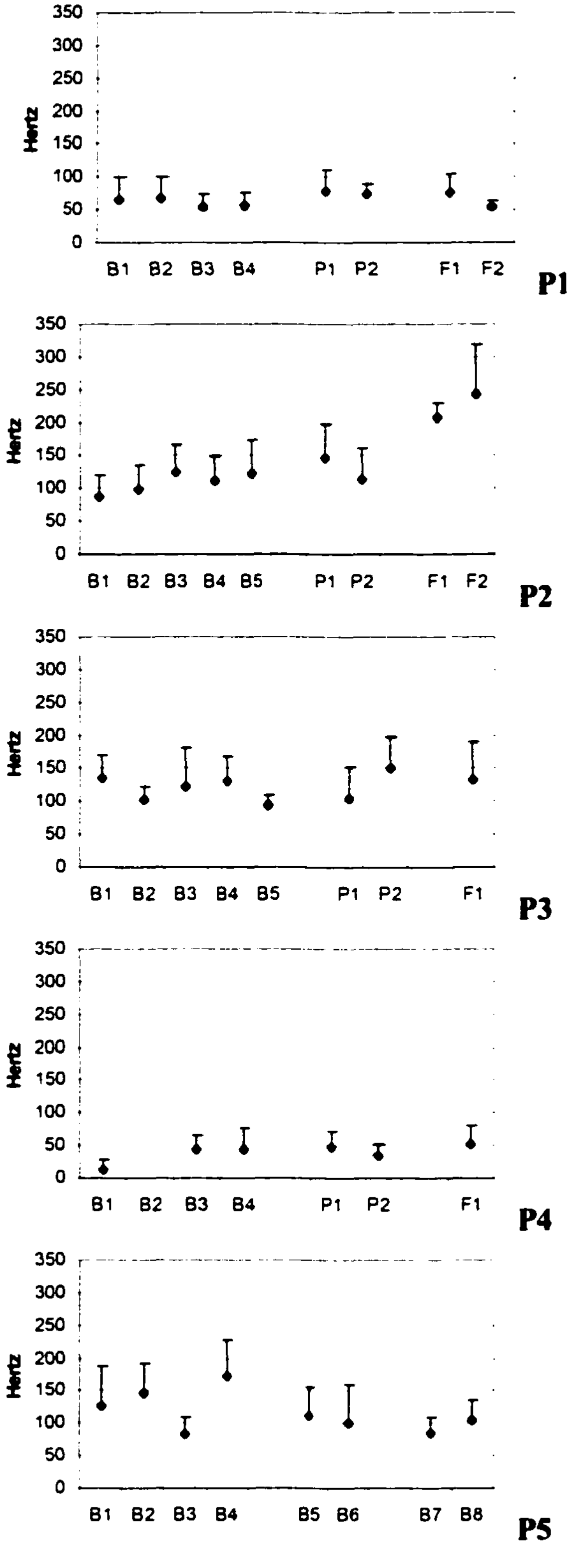


Figure 10. Percentage of listener preference for "overall loudness" of speech samples across study phases for each participant (P1-P5). Bars in the graph represent the percentage of preference for speech samples from Base, Post, and FUP. Percentage scores for a rating of "no preference" are not displayed in the graphs, but can be inferred by the degree to which the graphed values add up to 100%.

Figure 10. Percentage of listener preference for "overall loudness."

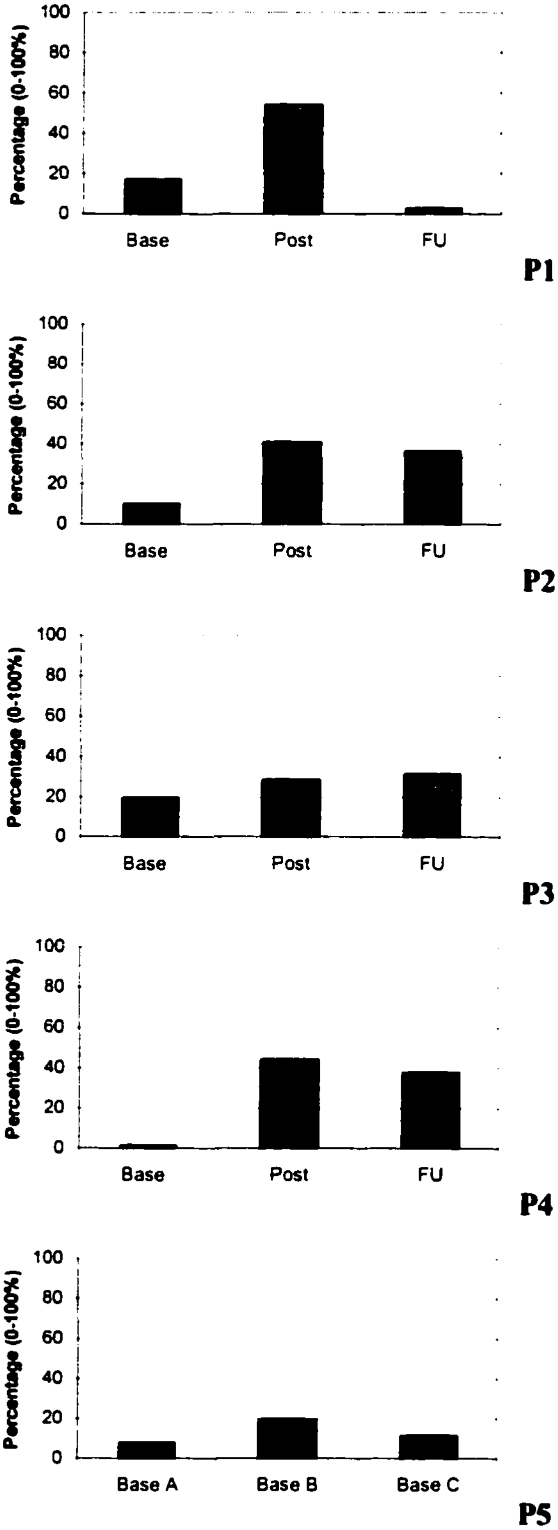


Figure 11. Percentage of listener preference for "loudness variability" of speech samples across study phases for each participant (P1-P5). Bars in the graph represent the percentage of preference for speech samples from Base, Post, and FUP. Percentage scores for a rating of "no preference" are not displayed in the graphs, but can be inferred by the degree to which the graphed values add up to 100%.

Figure 11. Percentage of listener preference for "loudness variability."

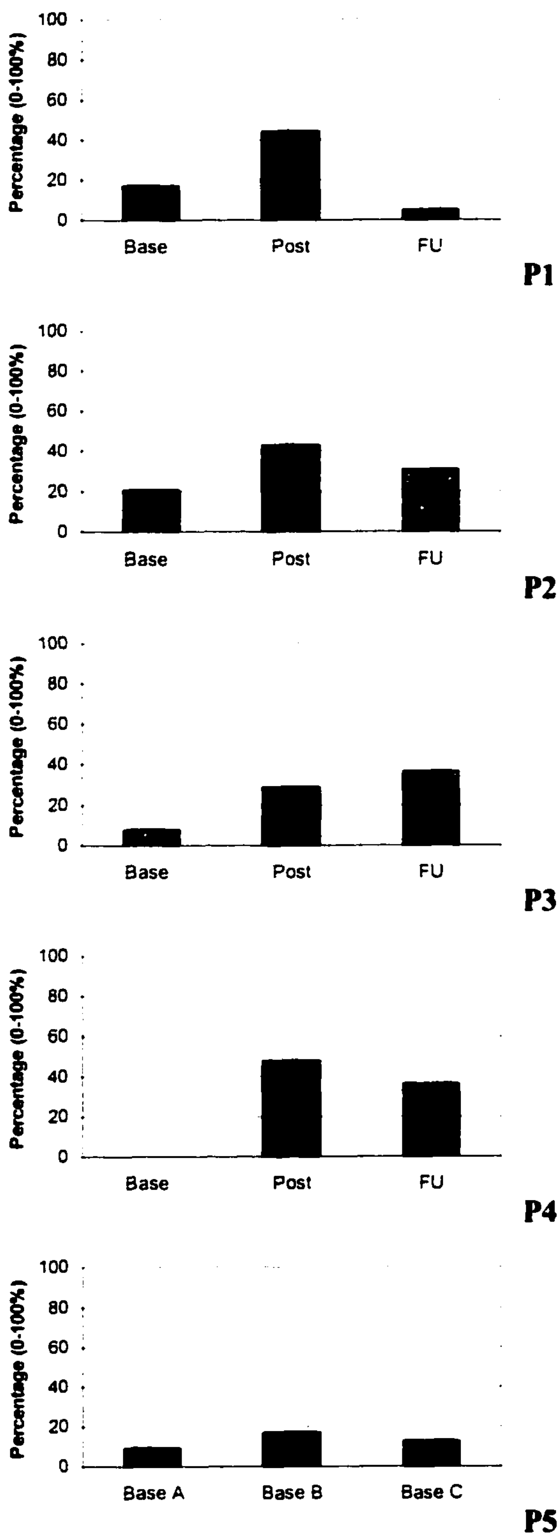


Figure 12. Percentage of listener preference for "overall pitch" of speech samples across study phases for each participant (P1-P5). Bars in the graph represent the percentage of preference for speech samples from Base, Post, and FUP. Percentage scores for a rating of "no preference" are not displayed in the graphs, but can be inferred by the degree to which the graphed values add up to 100%.

Figure 12. Percentage of listener preference for "overall pitch."

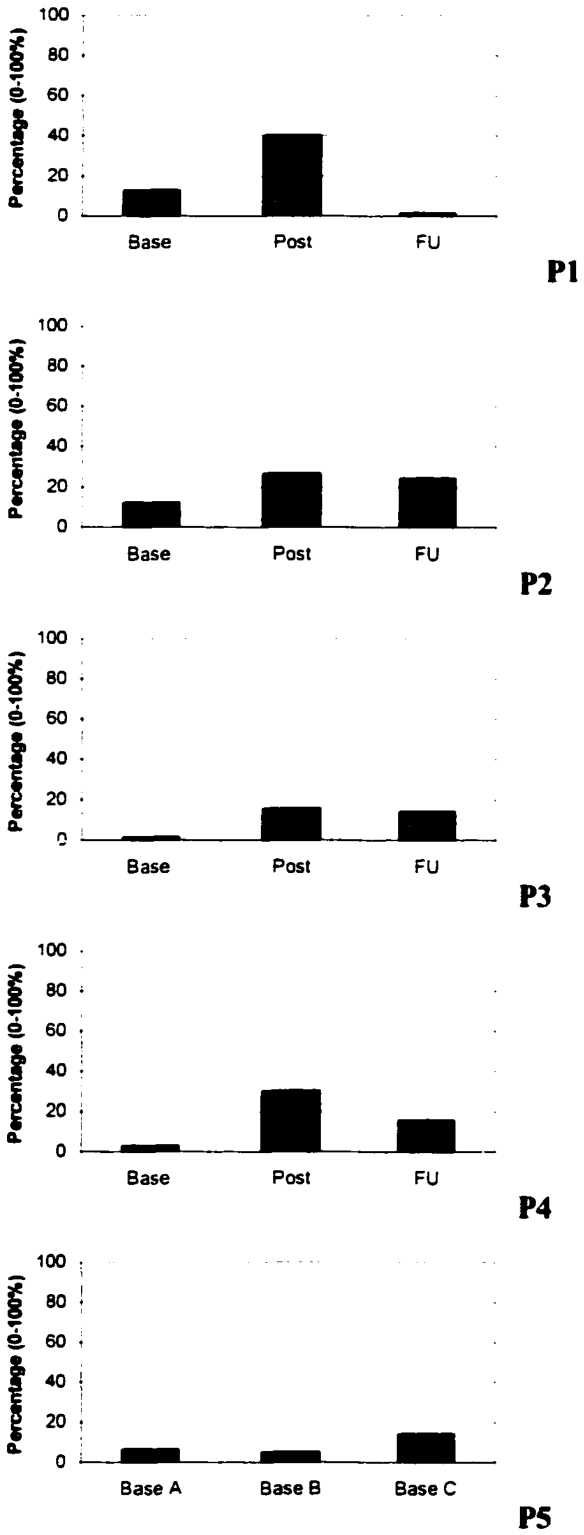


Figure 13. Percentage of listener preference for "pitch variability" of speech samples across study phases for each participant (P1-P5). Bars in the graph represent the percentage of preference for speech samples from Base, Post, and FUP. Percentage scores for a rating of "no preference" are not displayed in the graphs, but can be inferred by the degree to which the graphed values add up to 100%.

Figure 13. Percentage of listener preference for "pitch variability."

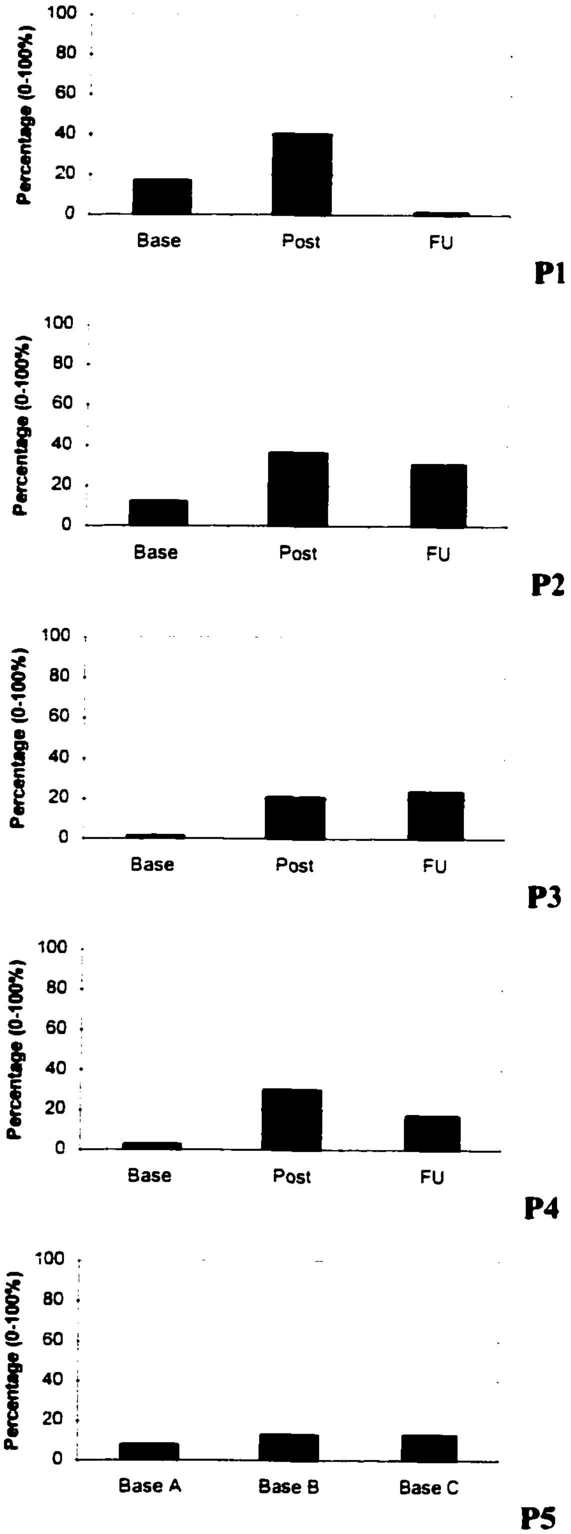
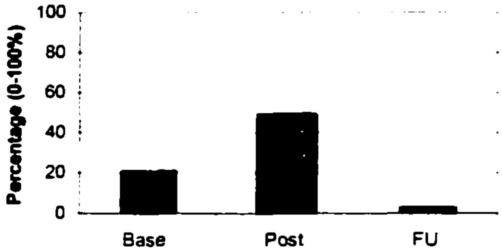
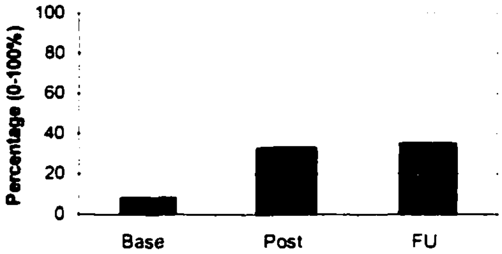


Figure 14. Percentage of listener preference for "overall voice quality" of speech samples across study phases for each participant (P1-P5). Bars in the graph represent the percentage of preference for speech samples from Base, Post, and FUP. Percentage scores for a rating of "no preference" are not displayed in the graphs, but can be inferred by the degree to which the graphed values add up to 100%.

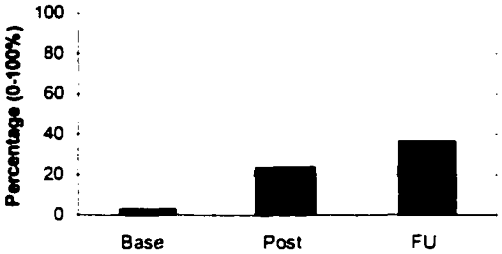
Figure 14. Percentage of listener preference for "overall voice quality."



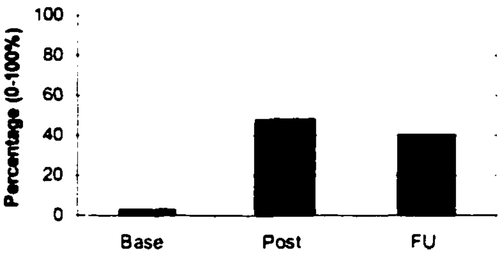
P1



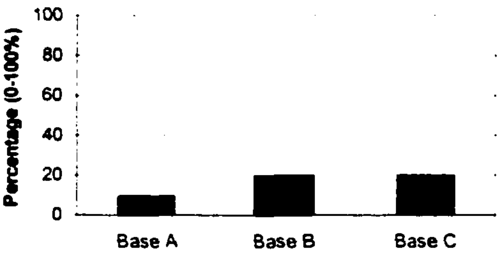
P2



P3



P4



P5

Figure 15. Percentage of listener preference for "articulatory precision" of speech samples across study phases for each participant (P1-P5). Bars in the graph represent the percentage of preference for speech samples from Base, Post, and FUP. Percentage scores for a rating of "no preference" are not displayed in the graphs, but can be inferred by the degree to which the graphed values add up to 100%.

Figure 15. Percentage of listener preference for "articulatory precision."

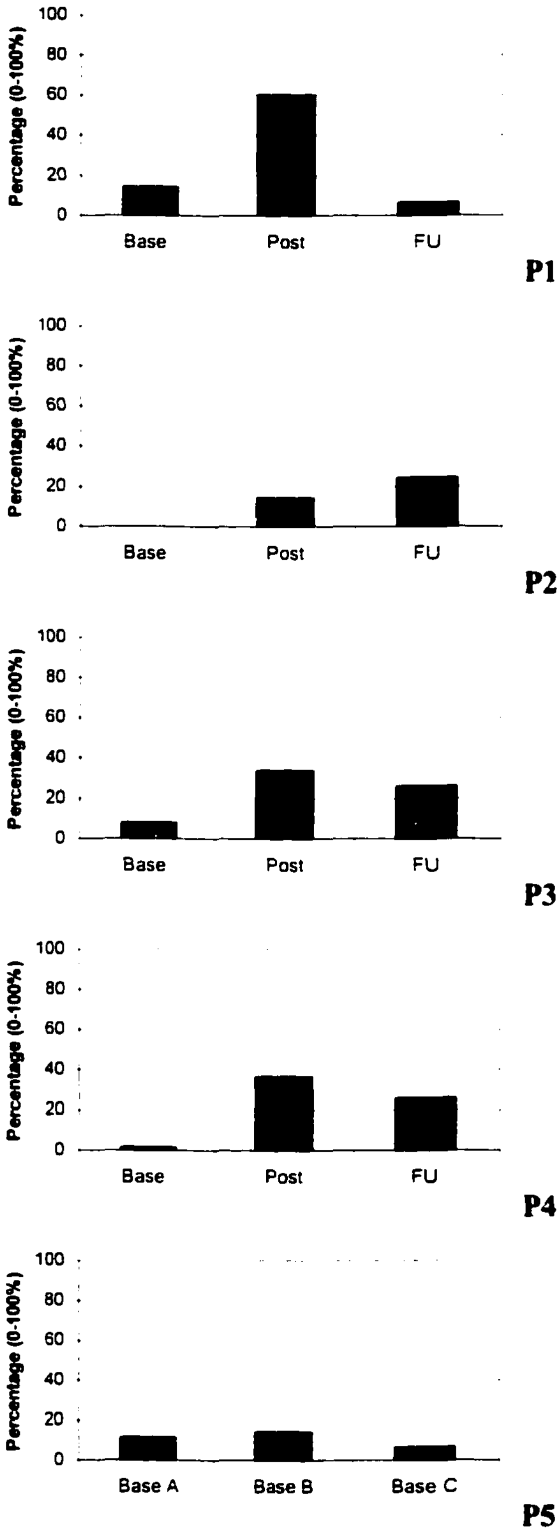


Figure 16. Percentage scores of parent perceptual ratings of voice, speech, and spoken communication characteristics of their children with CP across study phases. The percentage scale ranges from 0-100% with lower percentage scores representing a more severely impaired parent perception of a voice, speech, or communication characteristic. The black bar represents Base ratings, the white bar represents Post ratings, and the gray bar represents FUP ratings.

Figure 16. Parent perceptual ratings of speech, voice and communication characteristics.

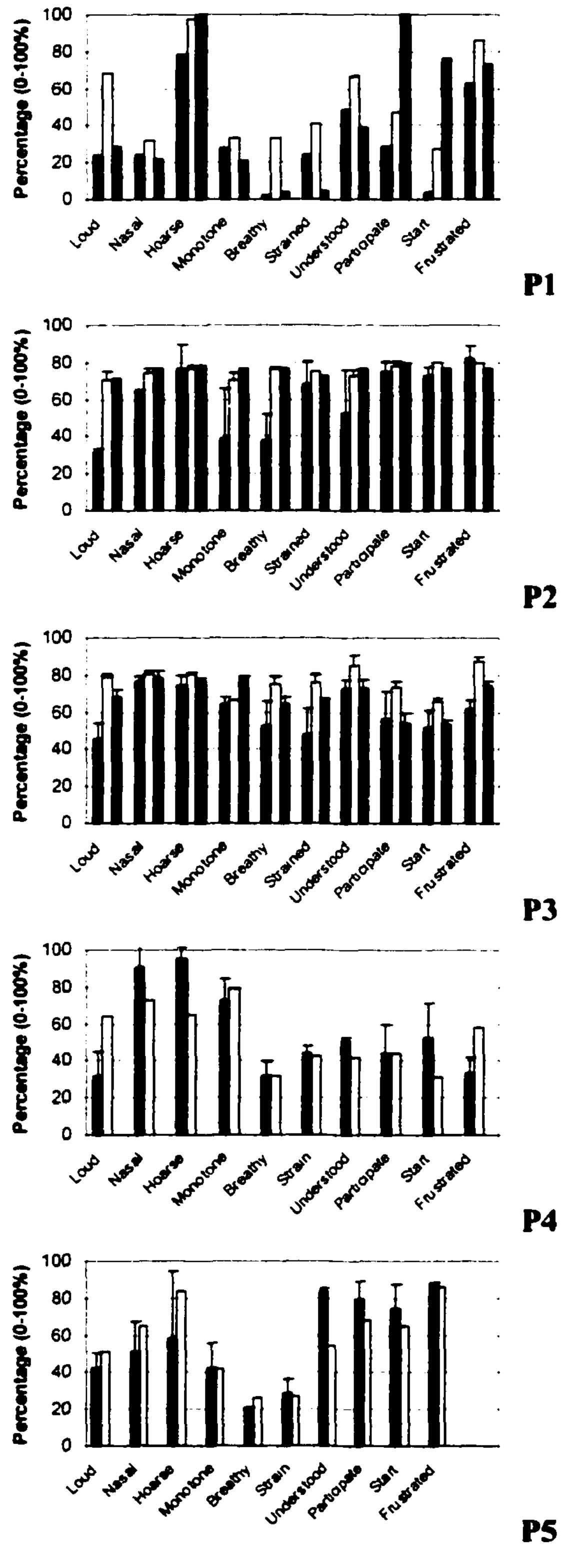


Table 1. Participant assignment to study conditions and actual number of recording and treatment sessions. The letter in parentheses (e.g., P1(A)) indicates the study condition to which the participant was randomly assigned. NC represents the age- and sex- matched typically developing peers. Each B_a represents a baseline recording session, each T represents a treatment session, each P_t represents a post-treatment recording session, and each F represents a follow-up recording session.

Weeks in Study

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16
P1 (A)	B_aB_a	B_aB_a	TTTT	TTTT	TTTT	TTTT	P_tP_t						FF			
P2 (C)	B_a	B_a	B_a	B_a	B_a	TTTT	TTTT	TTTT	TTTT	P_tP_t						FF
P3 (B)	B_a	B_a	B_aB_a B_a	TTTT	TTTT	TTTT	TTTT	P_tP_t						FF		
P4 (A)	B_a	B_aB_a	B_a	TTTT	TTTT	TTTT	TTTT	P_tP_t						FF		
P5 (D)	B_aB_a		B_aB_a					B_aB_a						B_a B_a		
NC1	B_aB_a															
NC2	B_aB_a															
NC3	B_aB_a															
NC4	B_aB_a															
NC5	B_aB_a															

Table 2. Grand means (and SDs) and mean differences for acoustic measures across tasks at Base, Post and FUP for P1. The arrows indicate the direction of change in performance between study phases (up arrow increased performance, down arrow decreased performance). The bold, thick arrows represent differences between study phases that were accompanied by non-overlapping values (strong trend). The thin arrows represent differences between study phases where values were overlapping. There are no arrows for the cartoon description task due to a single data point per acoustic measure at Base, Post, and FUP.

Table 2. Grand means (and SDs) and mean differences for P1.

Maximum Performance Tasks

MEASURES	Base	Post	FU	Base - Post	Base - FUP
Maximum vowel Duration (sec)	2.4 (1.0)	3.5 (1.0)	4.4 (0.5)	1.1 ↑	2.0 ▲
Maximum f_0 (Hz)	425 (44)	607 (177)	515 (59)	182 ▲	90 ▲
Minimum f_0 (Hz)	225 (38)	171 (38)	215 (15)	-54 ▲	-10 ↑
Maximum f_0 Range (Hz)	200 (42)	436 (139)	301 (45)	236 ▲	101 ▲

Sustained Phonation - three longest vowels

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	87.4 (4.9)	87.4 (2.9)	87.4 (7.2)	0	0
f_0 (Hz)	308 (33)	290 (6)	279 (45)	-18	-29
HNR (decibel)	22.1 (2.3)	22.9 (0.7)	24.1 (0.8)	0.8 ↑	2.0 ↑

Sentence Repetition

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	75.0 (1.6)	76.4 (1.3)	76.0 (0.7)	1.4 ↑	1.0 ↑
f_0 (Hz)	272 (7.6)	263 (15.5)	266 (0.9)	-9.0	-6.0
f_0 Range (Hz)	62 (7.0)	76 (3.1)	66 (16.2)	14.0 ▲	4.0 ↑

Cartoon Description

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	77.8 (6.5)	77.7 (4.5)	80.4 (5.0)	-0.1	2.6
f_0 (Hz)	304 (41)	277 (21)	333 (54)	-27.0	29.0
f_0 Range (Hz)	92 (36)	95 (61)	130 (72)	3.0	38.0

Table 3. Grand means (and SDs) and mean differences for acoustic measures across tasks at Base, Post and FUP for P2. The arrows indicate the direction of change in performance between study phases (up arrow increased performance, down arrow decreased performance). The bold, thick arrows represent differences between study phases that were accompanied by non-overlapping values (strong trend). The thin arrows represent differences between study phases where values were overlapping. There are no arrows for the cartoon description task due to a single data point per acoustic measure at Base, Post, and FUP.

Table 3. Grand means (and SDs) and mean differences for P2.

Maximum Performance Tasks

MEASURES	Base	Post	FU	Base - Post	Base - FUP
Maximum Duration (sec)	1.8 (0.6)	5.7 (0.4)	7.0 (1.4)	3.9 ▲	5.2 ▲
Maximum f_0 (Hz)	593 (261)	997 (163)	1323 (18)	404 ↑	730 ▲
Minimum f_0 (Hz)	304 (46)	273 (16)	239	-31 ↑	-65 ▲
Maximum f_0 Range (Hz)	289 (215)	724 (147)	1097	435 ▲	808 ▲

Sustained Phonation – three longest vowels

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	86.3 (1.5)	94.6 (1.1)	99.4 (1.0)	8.3 ▲	13.1 ▲
f_0 (Hz)	346 (13)	383 (7)	502 (37)	37 ▲	156 ▲
HNR (decibel)	22.0 (2.3)	22.0 (2.8)	24.0 (1.3)	0.0	2.2 ↑

Sentence Repetition

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	73.0 (1.9)	82.0 (0.4)	83.5 (4.4)	8.9 ▲	10.4 ▲
f_0 (HZ)	294 (11)	354 (10)	380 (3)	60 ▲	86 ▲
f_0 Range (Hz)	109 (16)	131 (23)	225 (26)	22 ↑	116 ▲

Cartoon Description

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	80.3 (3.4)	93.5 (4.8)	86.1 (2.4)	13.2	5.8
f_0 (Hz)	320 (35)	486 (78)	383 (34)	166	62
f_0 Range (Hz)	168 (85)	278 (101)	239 (81)	110	71

Table 4. Grand means (and SDs) and mean differences for acoustic measures across tasks at Base, Post and FUP for P3. The arrows indicate the direction of change in performance between study phases (up arrow increased performance, down arrow decreased performance). The bold, thick arrows represent differences between study phases that were accompanied by non-overlapping values (strong trend). The thin arrows represent differences between study phases where values were overlapping. There are no arrows for the cartoon description task due to a single data point per acoustic measure at Base, Post, and FUP.

Table 4. Grand means (and SDs) and mean differences for P3.

Maximum Performance Tasks

MEASURES	Base	Post	FU	Base - Post	Base - FUP
Maximum Duration (sec)	5.3 (0.8)	6.8 (0.6)	13.4	1.5 ↑	8.1 ▲
Maximum f_0 (Hz)	986 (163)	783 (103)	1160	-204 ↓	174 ↑
Minimum f_0 (Hz)	190 (38)	150 (9)	155	-40.5 ▲	-35 ▲
Maximum f_0 Range (Hz)	796 (127)	633 (112)	1005	-163 ↓	209 ↑

Sustained Phonation – three longest vowels

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	82.4 (2.7)	84.2 (2.8)	87.2	1.8 ↑	4.8 ▲
f_0 (Hz)	306 (44)	284 (19)	294	-23 ↓	-12 ↓
HNR (decibel)	19.2 (4.9)	21.7 (0.5)	23.3	2.5 ↑	4.1 ↑

Sentence Repetition

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	80.4 (2.9)	79.3 (2.9)	82.9	-1.13 ↓	2.5 ↑
f_0 (HZ)	261 (16)	266 (18)	269	4	7
f_0 Range (Hz)	118 (18)	128 (32)	134	10 ↑	17 ↑

Cartoon Description

MEASURES	Base	Post	FU	Base - Post	Base - FUP
SPL (dB SPL)	87.9 (4.8)	80.5 (4.0)		-7.4	
f_0 (Hz)	380 (93)	319 (36)		-60	
f_0 Range (Hz)	263 (125)	205 (81)		-58	

Table 5. Grand means (and SDs) and mean differences for acoustic measures across tasks at Base, Post and FUP for P4. The arrows indicate the direction of change in performance between study phases (up arrow increased performance, down arrow decreased performance). The bold, thick arrows represent differences between study phases that were accompanied by non-overlapping values (strong trend). The thin arrows represent differences between study phases where values were overlapping. There are no arrows for the cartoon description task due to a single data point per acoustic measure at Base, Post, and FUP.

Table 5. Grand means (and SDs) and mean differences for P4.

MEASURES	Base	Post	FU	Base - Post	Base - FUP
Maximum Performance Tasks					
Maximum Duration (sec)	1.0 (0.7)	1.2 (0.2)	1.5 (0.4)	0.2↑	0.5↑
Maximum f_0 (Hz)	364 (35)	424 (13)	419 (12)	60 ▲	55 ▲
Minimum f_0 (Hz)	233 (11)	213 (35)	234 (30)	-20↑	11↓
Maximum f_0 Range (Hz)	137 (34)	211 (21)	185 (18)	74 ▲	47 ▲
Sustained Phonation – three longest vowels					
SPL (dB SPL)	83.1 (10.1)	87.5 (2.5)	89.4 (1.4)	4.3↑	6.3↑
f_0 (Hz)	302 (21)	314 (8)	324 (18)	12↑	22↑
HNR (decibel)	15.6 (4.4)	19.3 (2.0)	24.5 (0.7)	3.7↑	8.9 ▲
Sentence Repetition					
SPL (dB SPL)	70.4 (4.4)	75.0 (0.7)	76.8	4.6↑	6.4
f_0 (HZ)	271 (11)	275 (38)	285	4↑	14↑
f_0 Range (Hz)	34 (18)	42 (9)	52	9↑	18 ▲

Table 6. Grand means (and SDs) and mean differences for acoustic measures across tasks at Base, Post and FUP for P5. The arrows indicate the direction of change in performance between study phases (up arrow increased performance, down arrow decreased performance). The bold, thick arrows represent differences between study phases that were accompanied by non-overlapping values (strong trend). The thin arrows represent differences between study phases where values were overlapping. There are no arrows for the cartoon description task due to a single data point per acoustic measure at Base, Post, and FUP.

Table 6. Grand means (and SDs) and mean differences for P5.

Maximum Performance Tasks

MEASURES	Base A	Base B	Base C	A - B	A - C
Maximum Duration (sec)	6.7 (1.4)	7.4 (1.3)	7.3 (0.8)	0.6↑	0.5↑
Maximum f_0 (Hz)	1083 (118)	777 (63)	969 (8)	-307 ▼	-114 ▼
Minimum f_0 (Hz)	224 (27)	258 (18)	251 (60)	34↓	27↓
Maximum f_0 Range (Hz)	860 (146)	519 (81)	719 (69)	-341 ▼	-141↓

Sustained Phonation – 3 Longest "ahs"

MEASURES	Base A	Base B	Base C	A - B	A - C
SPL (dB SPL)	95.1 (1.4)	91.2 (5.0)	95.3 (4.2)	-3.9↓	0.2
f_0 (Hz)	369 (18)	375 (19)	362 (18)	5.8↑	-7.3↓
HNR (decibel)	22.6 (4.7)	22.9 (0.5)	21.7 (2.4)	0.3↑	-0.85↓

Sentence Repetition

MEASURES	Base A	Base B	Base C	A - B	A - C
SPL (dB SPL)	78.9 (2.4)	78.5 (1.9)	78.4 (2.5)	-0.4↓	-0.5↓
f_0 (HZ)	360 (14)	362 (1)	347 (3)	2.0↑	-13.0↓
f_0 Range (Hz)	132 (38)	106 (8)	96 (14)	-26↓	-36↓

Cartoon Description

MEASURES	Base A	Base B	Base C	A - B	A - C
SPL (dB SPL)	77.3 (2.9)	75.5 (1.2)	77.4 (2.6)	-1.7	0.2
f_0 (Hz)	364 (31)	355 (18)	347 (27)	-8.9	-16.6
f_0 Range (Hz)	202 (95)	216 (78)	145 (91)	14.1	-57.0

APPENDIX A

Screening Questionnaire for Potential Participants
Intensive voice treatment for children with cerebral palsy.

General Information

Date of contact: ____

Parents Names: ____

Child's Name: ____

Address: ____

____

Phone: ______________________________

Date of Birth: ______________________ Age: ______________

Sex: _______ M _______ F

Medical Information on Child

Diagnosis: ____

Date of Diagnosis: ____

Medications: ____

____

____

____

What are your child's current symptoms associated with the above diagnosis? ________

____

Other Medical Problems: ____

____

Neurologist: ____

Primary Physician: ____

Has your child ever had any of the following?

Head injury	yes _________	no _________
Stroke	yes _________	no _________
Neurosurgery	yes _________	no _________
Respiratory disease	yes _________	no _________
Head / neck surgery	yes _________	no _________

Speech/Language/Hearing Information

What are your child's current speech symptoms, if any? ________________

____

____

Do people ever ask your child to repeat? ____________________________

____

____

What is your biggest concern regarding your child's communication abilities? _________

____

Has your child had, or is your child currently enrolled in speech therapy? ______

____

____

If so, what kinds of exercises has your child done, or does your child do, in speech therapy?

____

____

Does your child have problems with hearing? ______________________________

____

____

<u>Social Information</u>

Who does your child communicate with on a daily basis? (family, friends, teachers) ____

____

____

Do you think your child is ever frustrated during attempts to communication? ________

____

____

<u>Comments:</u>

APPENDIX B

Initial Interview – Child
Intensive voice treatment for children with cerebral palsy.

Subject code: ______________________

Session: ______________________ **Date:** ______________________

1. What kinds of talking do you do everyday? (who do you talk to, when do you talk most)

2. Do you like talking with other kids? Teachers? Siblings? Parents?

3. Is there any time when it is easier/harder for you to talk?

4. What do you do to make your talking sounds its "best"? (e.g. If someone doesn't understand what you say, is there anything you can do to make your talking sound better?)

5. When is it most important for you to have good "talking"? (e.g. when talking at school, home, during activities)

6. Can people always understand your talking? (kids, teachers, siblings, parents)

7. What do you like best about your talking? Is there anything you would like to change about your talking?

8. What kinds of games/activities do you like to do?

Other comments:

APPENDIX C

Initial Interview – Parent
Intensive voice treatment for children with cerebral palsy.

Subject code: ____________________

Session: ____________________ **Date:** ____________________

1. Tell me about your child's communication skills.

2. What kinds of talking does your child do on a daily basis?

3. How do you think your child feels about talking with other children? Teachers? Siblings? Parents?

4. Is there any time when it is easier/harder for your child to talk?

5. Is there anything you have figured out that you or your child can do to make his/her talking better?

6. Can people always understand your child? (children, teachers, siblings, parents)

7. Do you have any concerns regarding your child's communication skills?

8. Has your child ever had speech therapy? If so, when, for how long, what kinds of exercises?

9. What kinds of extracurricular activities does your child participate in?

10. Would you characterize your child as being high/medium/low energy level on a daily basis?

11. What are your child's favorite games, toys, sports, music stars, etc?

Post-treatment Questions

1. Did you notice any changes in your child's speech or voice functioning as a result of speech therapy?
   Describe:

2. Has anyone else notice any changes in your child's speech or voice?
   Describe:

3. What kinds of practicing does your child do?

   Do you think it helps?

4. Do people have a hard time understanding your child now?
   Describe:

5. Is there anything your child has learned to do that makes it easier for people to understand him/her?

6. Does your child do any more talking now after speech therapy?

7. Do people ask your child to repeat?

8. What did you like or not like about the speech therapy that your child received?

9. Would you recommend this treatment for other children with cerebral palsy?

APPENDIX D

VOICE RECORDING SESSION PROTOCOL

Subject Code: ______________________

Date of Recording: ______________________ **Time of Day**: __________

Last Medication: __________________ **Next Medication**: _________________

Human Subject Form: ______________

Perceptual Forms to Parents: ______________

Set – Up:

Sound level meter	_____	Video Camera	_____
SLM Extension cord	_____	Video microphone	_____
SLM Microphone Stand	_____	TV/VCR	_____
DAT Recorder	_____	DAT Tapes	_____
DAT Microphone	_____	Videotape	_____
Amplifier	_____	Stop Watch	_____
Korg Tuner	_____	Ruler	_____
Korg Microphone	_____	Tape for head microphone	_____

Speech Stimuli: Weiss Intelligibility test, Picture for description, Cartoon video

CALIBRATION:______________________________dB SPL at 4 inches

VOICE RECORDING:

Tape microphone to child's forehead. BE SURE DISTANCE IS EXACTLY 4 INCHES AND MICROPHONE IS WELL-SECURED!!!!

In the booth:

FOREHEAD MIC – ON________

VIDEO CAMERA MIC – ON__________

VIDEO CAMER – ON ________
(Hit play and record – make sure numbers are counting)

Outside the booth:

DAT RECORDER – ON________
(Hit play and record – make sure numbers are counting)

Monitor voice and video on TV screen

________________	DAT Rec. Level
________________	Microphone Distance
________________	Sound Level Meter Distance
________________	Pre-amplifier Level
________________	Camera to Forehead Distance

FATAL FLAWS:
- **Mouth to DAT microphone distance (4 inches)**
- **Mouth to SLM microphone distance (30 cm) – Measure constantly**
- **Always model with normal pitch and loudness – minimize behavioral cues with the exception of maximum performance tasks**

START:
State out loud on the
1.) DATE,
2.) Time, and
3.) Subject Code

TASK: MAXIMUM DURATION SUSTAINED VOWEL PHONATION "AH"

Directions: We are going to play a game to see how long you can say the sound "ah". The goal is to say "ah" for as *long* as you possibly can. OK, let me show you how to do it (Demonstrate Long "ah" at normal pitch and normal loudness). Now it is your turn.
Data Collector: Stimulate task from child
Data Monitor: Record peak dB SPL numbers at 1-sec. intervals from the SLM, record duration with stopwatch

Vowel 1: "Take a deep breath and say "ah" as long as you can."

____

____ Duration: ______ sec.

Vowel 2: Let's try it again and see if you can go even *longer*. Take a deep breath and say 'ah' as long as you can."

____

____ Duration: ______ sec.

Vowel 3: "See if you can go even *longer*.

____

____ Duration: ______ sec.

Vowel 4: *Longer…* Take a deep breath and say 'ah' as long as you can."

____

____ Duration: ______ sec.

Vowel 5: Take a deep breath and say 'ah' as long as you can."

____

____ Duration: ______ sec.

Vowel 6: "Take a deep breath and say 'ah' as long as you can."

____

____ **Duration: ______ sec.**

TASK: MAXIMUM PHONATION FREQUENCY RANGE

Data Collector: Stimulate task from child
Data Monitor: Record octave and musical note from the Korg Tuner
Directions: We are going to play a game with the sound "ah". This time we are going to say "ah" as high as we can and as low as we can. Let me show you how to play the game (Demonstrate a high "ah" and a low "ah"). Now you try one (Practice a high and a low). OK, when you are ready, take a breath and say "ah" going high.

High 1 : "Take a deep breath and say "ah" as high as you can."

Octave: ________________ Note: ________________

High 2: Let's try it again and see if you can go even *higher*. Take a deep breath and say 'ah' as high as you can."

Octave: ________________ Note: ________________

High 3: *Higher…* Take a deep breath and say 'ah' as high as you can."

Octave: ________________ Note: ________________

High 4: "Take a deep breath and say 'ah' as high as you can."

Octave: ________________ Note: ________________

Low 1 : "Now, when you are ready Take a deep breath and say "ah" as low as you can."

Octave: ________________ Note: ________________

Low 2: "Let's try it again and see if you can go even *lower*. Take a deep breath and say 'ah' as low as you can."

Octave: ________________ Note: ________________

Low 3: "Take a deep breath and say 'ah' as low as you can."

Octave: ________________ Note: ________________

Low 4: "Can go even *lower*. Take a deep breath and say 'ah' as low as you can."

Octave: ________________ Note: ________________

TASK: INTELLIGIBILITY SENTENCES

Data Collector: Stimulate task from child. MODEL – with normal pitch and loudness
Data Monitor: Record peak dB SPL numbers at 1-sec. intervals from the SLM

Directions: "I am going to say some sentences. After I say the sentence, I will point to you and I want you to repeat what I say. Let's practice (Say sentence, have child repeat). Nice job. OK, let's begin. Remember I will say the sentence first. Wait until I point to you, and then you repeat what I said."

"Buy Bobby a puppy"

___

"The potato stew is in the pot"

___

"The blue spot is on the key"

___

"Buy Bobby a puppy"

___

"The potato stew is in the pot"

___

"The blue spot is on the key"

___

"Buy Bobby a puppy"

___

"The potato stew is in the pot"

___

"The blue spot is on the key"

___

<u>TASK: DISCOURSE TASK – CANARY ROW:</u>

<u>Data Collector</u>: Stimulate task from child
<u>Data Monitor</u>: Record peak dB SPL numbers at 1-sec. intervals from the SLM

<u>Directions:</u> "I am going to show you a cartoon. It will last about three minutes. After you have seen the cartoon, you will tell (NAME) the story of the cartoon. After that, (NAME) will tell the story to someone else. She will not get to see the cartoon, so it is very important for you to tell her the story clearly and completely, so that she will be able to tell the story to someone else. "

<u>Comments:</u>

Parent/Caregiver Post-recording interview:

1. Was your child's speech and voice behavior today

Usual Better than Usual Worse than Usual

Comments:

2. Was your child's motivation/mood for participating today

Usual Better than Usual Worse than Usual

Comments:

3. Was your child's alertness/wakefulness today

Usual Better than Usual Worse than Usual

Comments:

CONCERNS FOR ANALYSIS:

Perceptual Rating Forms

Data Collector ___________

Data Monitor ___________

END CALIBRATION TONE: ________________________ dB SPL at 4 inches

APPENDIX E

Perceptual Rating Form

Code: ____________ **Date:** ______ **Relation to Child:** ____________________

Please mark the place on the line that best represents the child's typical speech:

Never loud enough enough	Always loud
Never a "nasal" voice	Always a "nasal voice"
Never a hoarse "scratchy" voice	Always a hoarse "scratchy" voice
Never monotone	Always monotone
Never a breathy voice	Always a breathy voice
Never a "strained" voice	Always a "strained" voice
Always speaks so others can understand	Never speaks so others can understand
Always talks when playing with kids	Never talks when playing with kids
Always starts talking/conversations with other kids	Never starts talking/conversations with other kids
Always frustrated when talking	Never frustrated when talking

APPENDIX F

Treatment Rating Form
(Adapted from the University of Iowa)
Intensive voice treatment for children with cerebral palsy.

1. How much do you think that your child's recent voice therapy improved your child's voice, overall?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

2. How much do you think your child liked the therapy approach that was used?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

3. How much did you like the therapy approach that was used?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

4. To what extent do you think therapy made your child's voice easier?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

5. To what extent do you think therapy made your child's voice clearer?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

6. To what extent do you think therapy made your child's voice louder?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

7. To what extent do you accept the type of voice use that was trained for your child in therapy?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

8. To what extent do you feel your child can be himself/herself, using his/her voice the way it was learned in therapy?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

9. How much fun do you think therapy was for your child?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

10. How <u>interesting</u> do you think therapy was for you child?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

11. To what extent do you think your child is technically able to use the technique trained in therapy in "real life" situations?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

12. To what extent do you think your child is <u>willing</u> to use the technique trained in therapy in "real life" situations?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

13. To what extent do you think your child will use the technique trained in therapy in "real life" situations in the future?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

14. To what degree would you recommend this type of therapy to a friend who has a child with cerebral palsy who is having speech and voice problems?

1	2	3	4	5
Not at all	Very little	Somewhat	Quite a bit	A lot

APPENDIX G

LISTENER TASK

Code:____________ Listener:__________________________

SAMPLES **Circle a choice or write in comments about WHY you preferred one sample to another**

Pair 1:

Overall Loudness:	A ☐	B ☐	No Preference ☐	Louder	Softer	Other	__________________
Loudness Variability:	A ☐	B ☐	No Preference ☐	More Variable	Less Variable	Other	__________________
Overall Pitch:	A ☐	B ☐	No Preference ☐	Higher	Lower	Other	__________________
Pitch Variability:	A ☐	B ☐	No Preference ☐	More Variable	Less Variable	Other	__________________

SAMPLES **COMMENTS**

Pair 1:

Overall Voice Quality:	A ☐	B ☐	No Preference ☐	Comments	______________
Articulatory Precision:	A ☐	B ☐	No Preference ☐	Comments	______________

APPENDIX H

Grand means (and SDs) and range of performance for acoustic measures across tasks for typically developing peers.

MEASURES	Mean (SD)	Range
Maximum Duration (sec)	15.2 (2.2)	11.4 – 16.9
Maximum f_0 (Hz)	1938 (843)	1062 - 3106
Minimum f_0 (Hz)	202 (30)	179 – 253
Maximum f_0 Range (Hz)	1737 (820)	884 - 2853
dB SPL of 3 longest ahs	95.9 (2.3)	93.9 – 99.7
Mean f_0 of 3 longest ahs	337 (61)	269 – 434
HNR of 3 longest ahs	27.3 (1.1)	26.3 – 29.0
dB SPL of sentences	78.1 (5.2)	74.4 – 86.9
Mean f_0 of Sentences	256 (23)	224 – 276
f_0 Range of Sentences	133 (37)	96 – 165
% Voiced of Sentences	65 (10)	49 – 74
dB SPL of Cartoon	78.6 (3.5)	75.8 – 84.3
Mean f_0 of Cartoon	255 (26)	217 – 291
f_0 Range of Cartoon	125 (50)	75 - 200

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